CLINICAL UTILITY OF RISK MARKERS FOR AGGRESSION AND DESTRUCTION IN CHILDREN WITH INTELLECTUAL DISABILITY

By

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Thesis Overview

This thesis is comprised of two volumes and is submitted by Charlie Bamford for the Clinical Psychology Doctorate at the University of Birmingham. Volume One presents the research component of the course and consists of three papers. The first paper is a meta-analytic review of risk markers relating to presence of self-injury, aggression and destruction in individuals with intellectual disability. The second paper is an empirical study exploring the clinical utility of risk markers for aggression and destruction in children with intellectual disability and develops an algorithm to predict current presence of these behaviours based on risk markers. The final paper is an executive summary that provides an accessible overview of the two preceding papers.

Volume Two of the thesis consists of five clinical practice reports that were completed over the course of the doctorate. The first describes the assessment and formulation of a 21-year old male's experience of anxiety and is formulated from cognitive and behavioural perspectives. The second is a service evaluation of Positive Behavioural Support Training for staff in a Learning Disability Service working with people displaying behaviour that challenges. The third report presents a single case experimental design to analyse the effect of a CBT intervention for social anxiety in a 38-year old male with personality disorder. The fourth details the assessment, formulation, intervention and evaluation of work conducted with a 70-year-old gentleman presenting with anxiety, later diagnosed with high-functioning autism/Asperger's. The final report presents an abstract of an oral presentation case study.

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CHAPTER 1

15 years on: A meta-analytic study of risk markers for self-injurious behaviour, aggression and destruction in individuals with intellectual disabilities

1.1 Abstract

Rationale: Since the previous synthesis of data pertaining to risk markers for individual behaviours that challenge (McClintock et al., 2003), there has been an additional wealth of research, however no additional synthesis. A comprehensive understanding of risk markers for self-injury, aggression and destruction in people with intellectual disabilities has the potential to facilitate the development of strategies to identify at-risk individuals earlier and devise more well-informed interventions. **Method:** A meta-analysis was conducted incorporating the data from the previous meta-analysis ranging from 1968 to 2002 (McClintock et al., 2003), as well as additional data published between 2002 and 2018, totalling 60 papers. **Results:** Results indicated that level of intellectual disability and presence of autism was significant risk markers for all three behaviours. Additional risk markers for self-injury included: fragile X syndrome, increased repetitive behaviour, Cornelia de Lange syndrome, residing in paid care, tuberous sclerosis complex, visual deficit, expressive communication deficit, Prader Willi syndrome, Angelman syndrome, overactivity, hearing deficit, mobility deficit, adaptive behaviour deficit, epilepsy and female gender. Down syndrome represented decreased risk for self-injury and aggression. Additional risk markers for aggression included: living in paid care, epilepsy, expressive communication deficit and Smith Magenis syndrome. **Conclusion:** This meta-analysis highlights that data pertaining to self-injury is far more comprehensive than for aggression or destruction. It emphasises the need for additional research within these areas, as well as within specific syndromes. As advancements are made in understanding risk markers for specific behaviours, these may be used to inform interventions or earlier identification of individuals at particularly increased risk.

1.2 Introduction

1.2.1 Background

Intellectual disability (ID) is defined as a chronic, severe condition that limits an individual's adaptive behaviour and intellectual functioning, with traits consistent with diagnosis identified before the age of four (World Health Organisation, 1992). Approximately one in 100 individuals with ID will show behaviour that is considered challenging at some point during their lives (Emerson et al., 2001) that will persist without treatment (Totsika & Hastings, 2009). Self-injury, aggression and destruction have been a particular focus of research as these behaviours are often severe, and predictive of detrimental outcomes. The presence of behaviours that challenge is associated with psychiatric hospitalisation, reactive physical intervention and reduced quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009; Mandell, 2008; Allen, Lower, Brophy & Moore, 2009). Moreover, presence of these behaviours increases parental stress, result in high staff burn out and exclusion from school settings (McIntyre, Blacher & Baker, 2002; Knapp et al., 2005). The Winterbourne View scandal resulted in increased commitment to reduced inpatient hospitalisation as a result of behaviours that challenge. The Transforming Care and Commissioning Steering Group implemented following this scandal, chaired by Stephen Bubb, reported in 2014 that emphasis should be placed on helping people return to their homes, specifically stating that hospitals are not homes for individuals with ID. To proactively reduce psychiatric hospitalisation, and to ameliorate the downstream consequences of behaviours that challenge to individuals and their wider social systems, there has been renewed interest in early intervention for behaviours that challenge (Cooper et al., 2014). Early intervention is predicated on understanding risk markers (correlates) associated with forms of behaviours that challenge. Such an understanding would help to identify those at greatest risk of developing behaviours that challenge and may also help to identify novel intervention targets.

The operant model of behaviour has been applied to self-injury, aggression and destruction within intervention approaches. As a result of this, interventions are often drawn from applied behavioural analysis (e.g., Gardner, 2002) and applied to social skill development (Carr, 1997) and parent training models (Matson, Mahan & Matson, 2009). This, approach, however, does not take into consideration whether interventions tailored towards specific behaviours will be more effective. Despite the significant social, economic and quality of life consequences of behaviours that challenge (Emerson et al., 2002; Fox & Emerson, 2002), there has been limited synthesis of the literature regarding key demographic, personal or behavioural variables that are associated with each of these behaviours that challenge. Moreover, consideration of risk for behaviours that challenge at a unitary category level, rather than the individual behaviours of self-injury, aggression and destruction, fails to isolate variables that may have specific influence on each form of behaviour (McClintock, Hall & Oliver, 2003), or recognise that individuals may engage in multiple different forms of behaviours that challenge (Matson et al. 2008) with different risk pathways.

1.2.2 Self-injury

The term 'self-injury' or 'self-injurious behaviour' (SIB) includes behaviours that are self-directed, and that have the potential to, or actually cause physical harm to the individual, such as biting oneself, pulling one's hair, head banging and scratching one's self (Rojahn et al., 2008; Emerson et al., 2001; Bodfish et al., 1995). Reports of the prevalence of self-injurious behaviour among individuals with ID are diverse, ranging from 4% to 24% (Cooper et al., 2009; Deb, Thomas & Bright, 2001). This heterogeneity in prevalence estimates is largely attributed to different definitions of self-injury, for example whether it results in tissue damage, and differences in who reported the incident of SIB (poor concordance is reported between parent and teacher reports of behaviours that challenge, e.g., Stadnick, Chlebowski & Brookman-Frazee, 2017). Moreover, many studies utilise small samples; heterogeneity in

smaller samples can have a more substantive effect on the reported prevalence estimate, leading to high variation. It is essential to synthesise data across studies to ensure the sources of heterogeneity, such as small samples and differing definitions of behaviours that challenge, are accounted for in interpretation.

In individual cohorts, a number of risk markers for SIB have been identified, including severity of ID (Chadwick et al., 2000; Holden & Gitlesen, 2006; McClintock, Hall & Oliver, 2003), physical health difficulties that result in pain (Symons, 2011; Richards et al., 2017), autistic spectrum disorder diagnosis (Richards, Oliver, Nelson & Moss, 2012; McClintock, Hall & Oliver, 2003) and repetitive or impulsive behaviours (Rojan, Matson, Naglieri & Mayville, 2004; Bradley, Summers, Wood & Bryson, 2004; Cooper et al., 2009). In addition, a range of genetic syndromes have been identified as associated with SIB including Prader Willi syndrome (Holland et al., 2003) and Cri du Chat syndrome (Collins & Cornish, 2002), Smith Magenis syndrome (Arron et al. 2011) and fragile X fyndrome (Eden et al., 2014). The identification of valid risk markers confers the opportunity to develop theoretical models pertaining to the mechanisms underpinning SIB. For example, operantly derived models of SIB indicate that SIB serves a particular function for an individual with ID. Functional analyses (Iwata et al., 1994) can help to determine the exact function, however commonly identified social functions include communication, escape, avoidance and access to tangibles (Carr, 1977). Thus, when an individual has limited expressive communication, which is a necessary feature of more severe ID, SIB may be more likely to form an effective functional 'communication' in that behaviours may be reinforced by the environment and become learnt. In contrast, there are also risk markers that may be associated with internal mechanisms, for example by sensory stimulation (Carr, 1977). Evidence suggests there could be a correlation between pain and increased self-injury, with it possibly moderating people's perception of pain (Wool & Salter, 2000). Moreover, some research indicates that genetic disorders correlated

with SIB may also have a higher threshold to pain (Priano et al., 2009). These putative unique associations between risk markers and SIB further strengthen the rationale to evaluate risk markers for each form of behaviour separately.

1.2.3 Aggression

'Aggression' can be used to refer to an array of acts, including physical assaults on peers, family members or staff, hostility or verbal threats, making gestures others consider threatening and having tantrums (Rojahn et al., 2001). Similarly to SIB, the prevalence of aggressive behaviour among individuals with ID varies considerably, ranging from 30% to 60% (Crocker et al., 2006; Cohen et al., 2010). The presence of aggressive behaviour presents a number of social difficulties for children with ID as it can result in exclusion from school, placements away from the family home, psychiatric intervention and difficulty socialising with peers (Allen, 2008; Lowe et al., 2007; Posey et al., 2008). Notably, Davies & Oliver (2016) indicate that aggression is the most persistent of the three forms of behaviours that challenge, with 69% of young children with ID displaying aggression. This suggests that there is significant value in understanding risk markers that make an individual more likely to display aggressive behaviour, as it may be particularly important to develop pre-emptive interventions, or better tailor current interventions to ameliorate this persistent behaviour.

Whilst a number of studies have postulated different risk markers for aggression, the results have been fairly contradictory. Some research indicates that severity of ID correlates with increased aggression (Meins, 1995; McTiernan et al., 2011), whilst alternative research indicates that higher ability evidenced through higher-level expressive communication is more closely associated with aggression (Emerson et al., 2001; Deb et al., 2001). Living accommodation has also been reported as a risk marker, with individuals living in their family home or alone being less inclined to act aggressively (Eyman et al., 1970). Moreover, studies considering specific syndromes linked with ID, such as Down syndrome (Collacott et al., 1998)

have considered whether the syndrome reduces or increases risk of aggression. Similarly to SIB, aggression has also been associated with overactivity/impulsivity (Davies & Oliver, 2016), as well as factors that cause internal pain, such as headaches or ear infections (Gardner & Whalen, 1996).

Some definitions of aggression encompass 'destruction of property'. Whilst this is an act of aggression, it is not comprehensively understood whether destruction of property should be considered separately due to being associated with different risk markers. It is an aim of this review to examine them separately and draw conclusions on this.

1.2.4 Destruction of property

To date, research has often considered 'destruction' under the umbrella of aggression, making it difficult to isolate the two forms of behaviour and consider risk markers associated with destruction specifically. Furthermore, the Behaviour Problem Inventory (BPI; Rojahn et al., 2001), which is a widely used measure to quantify behaviour that challenges, has subsections for SIB and aggression only; destruction of property forms part of the aggression subheading. This means that studies using this measure are unable to independently quantify prevalence and risk markers for this third subset of behaviours that challenge. For example, Croker et al. (2006) compared different forms of aggressive behaviour (verbal, property, physical, self and sexual). This indicated verbal aggression was considerably higher in males than property aggression and also showed that individuals with mild ID were more likely to show aggression than property destruction. These, and other divergent prevalence figures suggest that it is necessary to consider aggression and destruction as separate components of behaviours that challenge, as they may have different risk markers. Whilst there is limited evidence on risk markers for destruction specifically due to the conflation of aggression and destruction, presence of autism and overactivity/impulsivity have been presented as possible risk markers (Davies, 2010), gender (Crocker et al., 2007) and severity of ID (Crocker et al., 2006).

1.2.5 Meta-analyses

A previous meta-analysis by McClintock, Hall and Oliver (2003) synthesised risk data for SIB, aggression and destruction of property. The analysis included data available prior to 2002 that assessed each of these components of behaviours that challenge. The literature identified enabled the authors to examine the relative risk of a number of characteristics and the presence of SIB (gender, degree of ID, autism, receptive communication and expressive communication), aggression (gender, degree of ID, autism and expressive communication) and destruction of property (autism). This meta-analysis indicated that there were limited data available focussing on these specific forms of behaviours that challenge. Over forty papers have been published in the past decade that include data on risk markers and therefore there is significant clinical and scientific value in replicating and extending the original meta-analysis of risk markers relating to aggression, destruction and SIB (McClintock et al., 2003).

Therefore, the present meta-analysis has the following aim: to synthesise and quantify data on a) demographic[1], b) diagnostic[2] and c) health and person/behavioural risk markers[3] for SIB, aggression and destruction in people with ID and subsequently explore how each risk marker impacts the corresponding behaviour.[4]

[1] Demographic risk markers: Gender, Level of ID, Adaptive behaviour, Living arrangements

[2] Diagnostic risk markers: Angelman syndrome, Autism, Cornelia de Lange syndrome, Down syndrome, fragile X syndrome, Prader Willi syndrome, Smith Magenis syndrome, Tuberous sclerosis complex

[3] Health/behavioural risk markers: Visual deficit, Hearing deficit, Mobility deficit, Epilepsy, Dental problems, Gastrointestinal problems, Skin problems, Expressive communication, Receptive communication, Overactivity/impulsivity, Repetitive behaviour

[4] Categorisations are pragmatic to allow segmentation of data and appraisal of each risk marker in turn

1.3 Method

As the present study aimed to extend the meta-analysis conducted by McClintock, Hall & Oliver (2003), the search terms and methods used by McClintock and colleagues were used as the basis for the current study.

1.3.1 Literature Search

A search of published literature was conducted on 21st December 2018 to obtain a comprehensive overview of research focussing on ID and topographies of behaviours that challenge. The terms in Table 1.1 were used to form the search criteria and identify literature pertaining to each of the areas. To combine the search terms, boolean operator 'OR' was used to ensure the search contained all the variations of the terms indicated in Table 1 (e.g., Learning disab* OR Intellectual disab* OR Mental retardation OR Learning difficult*). Following this, the Boolean operator 'AND' was used to combine the behaviour search terms with intellectual disability search terms. These search terms were applied to the electronic databases: Ovid MedLine, Embase and Psyc Info. All search terms used (apart from 'behav* that challenges') were consistent with those used by McClintock and colleagues (2003). 'Behav* that challenges' was included due to progressive change in how 'challenging behaviour' is referred to; more recent papers may be more inclined to use this terminology. Attempts were not made to identify studies that were unpublished at the time of searching. This decision was made in line with the methodology used in McClintock et al. (2003). Whilst it is appreciated that this presents a risk of bias towards positive results, as these are more commonly published, only 30% of meta analyses include unpublished data as there is a rationale that it hasn't been peer reviewed and may be of a lower standard (Cook et al., 2003). Duplicates were removed during the electronic database search. As McClintock et al. (2003) ceased their search in late October 2001, the current search was limited to papers published between October 2001 to December 2018; to avoid excluding any studies published late in October 2001, this one-month overlap

was considered beneficial. The decision was made to continue the search from the cessation of the previous analysis, rather than repeating the previous search. Updating is considered more efficient than starting afresh when new evidence emerges (Garner et al., 2016). The previous review had clear and appropriate methods and search terms (that have not been significantly adjusted in the current analysis – only one addition has been made to represent progression in language use) and therefore this was deemed appropriate. Thus, the search was conducted focussing on papers published between October 2001 and 21st December.

Table 2.1 Search terms used to identify relevant papers

Search Term	Variations
Intellectual Disability	Learning disab*; Intellectual disab*; Mental retardation; Learning difficult*
Destruction	Destruction of property; Destructive behav*
Aggression	Aggression; Aggressive behav*
Self-Injurious Behaviour	Self-injury; Self-harm
Behaviour that challenges	Challenging behav*; Aberrant behav*; Maladaptive behav*; Problem behav*; Behav* problem; Behav* disorder; Behav* disturbance; Disturbed behav*; Behav* that challenges

1.3.2 Inclusion criteria

Table 1.2 provides a comprehensive overview of the inclusion/exclusion criteria during the title and abstract screening stage, and Table 1.3 indicates additional criteria applied during the full text search. The main criteria in the initial stage were reference to the presence of ID and reference to behaviour that challenges of any kind. Restrictions were later introduced during the full text search to ensure that sufficient data were available to calculate an effect size for each risk marker.

1.3.2.1 Title and abstract Screening

The use of the search terms indicated in Table 1.1 resulted in identification of 9985 papers. An initial title and abstract screen was conducted based on the inclusion/exclusion criteria outlined in Table 1.2.

Table 1.2 Inclusion/exclusion criteria for title and abstract screening

Inclusion	**Exclusion**
Publication date Oct. 2001 onwards	Publication prior to Oct. 2001
Reference to ID	No reference to ID
Reference to behaviour that challenges	No reference to behaviour that challenges
Empirical paper	Not an empirical paper (e.g., review)
Quantitative design	Qualitative design

1.3.2.2 Full text Screening

107 papers met the title and abstract inclusion criteria. A full text screen was subsequently conducted for each of these 107 papers, following the inclusion/exclusion criteria outlined in Table 1.3. In order to quantify the magnitude of risk associated with each risk marker, papers were only included where data were presented which would allow an effect size to be derived. In line with McClintock et al. (2003), to quantify the risk associated with a particular diagnosis (e.g., autism or a genetic syndrome), the paper needed to report data on the group of interest *and* a relevant control group. To reduce error, a sample size of 25 or greater for each group was imposed to avoid bias caused by sampling error in the meta-analysis and interpretation of within study variance as actual variance (Lin, 2018). Lin (2018) suggests that samples of 50 participants still present a risk of bias, however the incidence of some rare syndromes makes it almost impossible to achieve large sample sizes, therefore a sample of 25 was used. Whilst this does increase the risk of sampling error, it also reduces the potential to explore heterogeneity

and important sub-groups may not be analysed; therefore, a decision to include data from samples of 25 or greater was an appropriately conservative balance.

Table 1.3 Inclusion/exclusion criteria for full text screen

Inclusion	**Exclusion**
Sufficient data available to calculate an odds ratio or effect size	Insufficient data available to calculate an odds ratio or effect size
Control population used in cases of a population with autism or genetic syndromes	No control population used in cases of a population with autism
Specific risk markers mentioned (e.g., age, genetic syndromes, gender, impulsivity)	No specific risk markers mentioned
ID specified for target population	ID not specified for target population
Specific behaviour that challenges mentioned (self-injury, aggression, destruction)	No specific behaviour that challenges mentioned
Empirical study	Not an empirical study (e.g., conference proceedings, review articles, books)
Population not recruited for a primary characteristic that would be a covariate (e.g., recruited due to having epilepsy)	Population recruited for a primary characteristic that would be a covariate (e.g., recruited due to having epilepsy)
Sample size >25	Sample size <25
Full text written or available in English	Full text not available in English
Unique sample	Sample not unique – reported elsewhere

1.3.3 Manual search

The citations outlined in the included papers from the database search were manually searched to identify any relevant papers that had not arisen during the database search. These papers were subjected to the same two-stage inclusion/exclusion screening processes.

1.3.4 Inclusion of McClintock, Hall & Oliver (2003) data

The 20 papers from the previously published McClintock, Hall & Oliver (2003) meta-analysis were included were included prior to the full text screen to provide a complete meta-analysis

of studies published up to December 2018 relating to risk markers for SIB, aggression and destruction among individuals with ID that meet the inclusion criteria. These papers underwent the same inclusion/exclusion criteria screening as the newly added papers. No papers were excluded.

1.3.5 Search Results

The results of the systematic search are presented in Figure 1.1. The initial search produced 11,241 articles, reduced to 9985 once duplicates were removed. Of the 107 full text articles examined, 38 were included in the study. The three most common reasons for exclusion were that a specific type of behaviour that challenges (self-injury, aggression or destruction of property) were not mentioned (n=29), there was insufficient data available to determine an effect size (n=15) and the article was not available in English (n=9). An additional 2 articles were added through the manual search process and the 20 McClintock, Hall & Oliver (2003) articles were included, equating to 60 studies suitable for meta-analysis. The search, screening and data extraction were conducted in line with the PRISMA (Moher et al., 2009) checklist.

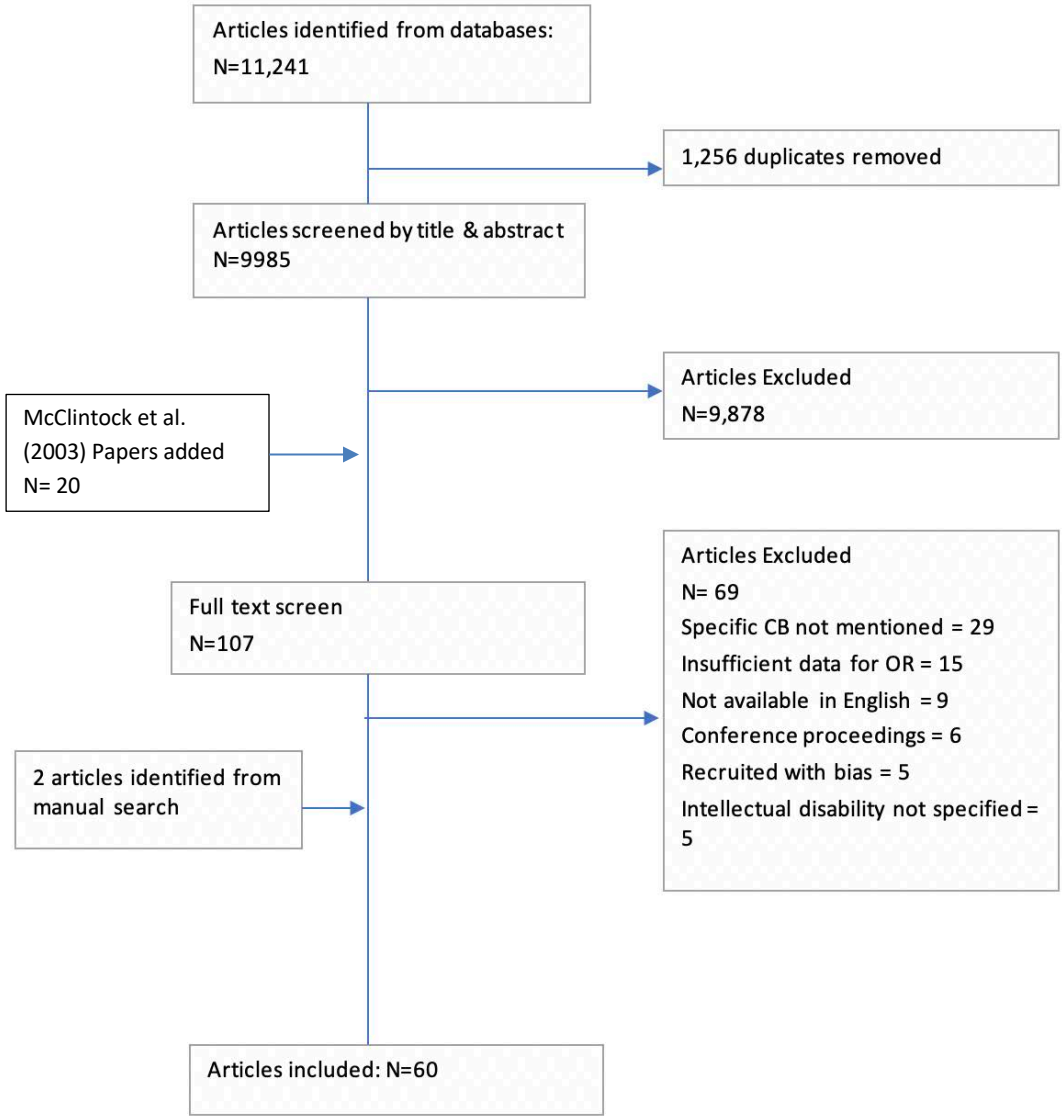

Figure 1.1: Results of systematic search and application of inclusion/exclusion criteria

1.3.6 Quality rating

The final 60 papers were rated for quality using the framework outlined in Table 1.4, adapted from Richards, Jones, Groves, Moss & Oliver (2015) and Surtees, Oliver, Jones, Evans & Richards (2018). The framework was developed based on factors considered to threaten the external and internal validity of the meta-analysis. Consistent with current diagnostic manuals

(e.g., DSM-IV), determination of ID was considered to have two distinct components; adaptive functioning and cognitive functioning (World Health Organisation, 1992). The reliability of quality ratings were cross-validated by a second rater using a 25% random sample. Total inter-rater reliability was 87% (based on the number of scores consistent out of total number scored). Discrepancies were reviewed and a consensus reached. The quality ratings were utilised for descriptive purpose and studies were not excluded or analysed on the basis of quality ratings in order to replicate the methodology adopted by McClintock, Hall & Oliver (2003).

Table 1.4 Quality framework, colour coded for ease of visual inspection

	0 Poor*	1 Adequate	2 Good	3 Excellent
Sample Identification	Unspecified	Single restricted/ non-random sample e.g. specialist clinic or single region	Multiple restricted/ non-random samples e.g. multi-region specialist clinics	Random or total population study
Measurement of Behaviour that Challenges	Unspecified	Staff/carer anecdotal report	Use of appropriate behaviour measure (e.g. Maladaptive Behaviour Checklist)	Direct observation or multiple appropriate behaviour measures
Measurement of Adaptive Functioning	Unspecified	Clinician judgement Self/parent report Recruited from a specialise ID school/support group	Self/parent report with well validated measure	Validated measure such as the Vineland Adaptive Behaviour Scales
Measurement of Intellectual Functioning	Unspecified	Syndrome group known to be associated with ID Self/parent report Recruited from specialist ID school/support group	Self/parent report with well validated measure	Formal IQ test (Wechsler Intelligence Scale for Children etc.)

**No studies met criteria for zero, therefore the lowest rating (1) was colour coded red for ease of visual inspection*

Of the studies included in the final analysis, 8.8% were considered adequate in terms of sample identification, 75.6% were good and 15.6% were excellent. In terms of measurement of behaviour that challenges, 15.5% used staff/carer anecdotal reports, 82.3% used an appropriate measure, whilst 2.2% observed the behaviour in question. 24.4% of included studies accepted participants with a known diagnosis as an indicator of adaptive behaviour difficulties, 60% used staff or carer reports and 15.6% were able to directly measure adaptive behaviour using a validated questionnaire/observation. Finally, 26.7% accepted a specified syndrome as indication that a deficit in intellectual functioning was present, 57.7% accepted parent or carer reports, and 15.6% assessed intellectual functioning with a validated measure. Given the time and cost restrictions of research, acceptance of a specific disorder as being known to be associated with intellectual disability or accepting parent/carer reports are the most common approaches to recruitment of individuals with intellectual disability. It is unlikely that parents would receive a diagnosis of intellectual disability without careful assessment from professionals. Whilst there is a concern that some participants included in samples may not be representative of the target population if they are not assessed by the research team, the means of inclusion do not raise considerable concern.

1.3.7 Data Extraction

In order to calculate relative risk (RR), event rates were reported as the number of participants with and without the condition of interest in both a control and exposure/risk group. Where event rates were not given, and RR was instead specified, this was included if associated confidence intervals for the RR were also provided. Authors were contacted to obtain original RR and confidence interval data to avoid using regression or where error bars were used to indicate confidence intervals, rather than exact numbers. In the case of multiple reporting of outcomes from one primary study, if it were possible, these were combined in a single quantitative outcome using the procedures described by Borenstein (2009). If it were not

possible to combine them, for example because they were separated into different age groups (e.g. Richards et al., 2017), each outcome was included. It should be noted that this method may result in a slight reduction in confidence intervals for the random effects model.

Where a paper considered a risk marker that was absent in the whole sample, the data were not extracted from this paper. For example, in the study conducted by Wilde and colleagues (2017), 100% of the participants had normal hearing, and thus it was not possible to include data from this paper on hearing deficits as a risk marker for behaviour as there was no event rate for hearing deficit to compare to those without. Where studies presented multiple forms of 'aggressive behaviour' (e.g., verbal, physical, self and sexual), physical aggression only was used, consistent with the data reported by McClintock et al. (2003). If studies presented information on 'aggressive-destructive' behaviour, which was a typical outcome for those using the Behaviour Problems Inventory (Rojahn et al., 2001), these data were excluded due to confounding two of the primary outcome variables confounds. This resulted in the exclusion of data from six studies that focussed on aggressive-destructive behaviour.

For risk markers that were not dichotomous (e.g. level of ID is usually categorised as profound severe, moderate and mild), data were grouped either in line with McClintock, Hall and Oliver (2003), or in the manner that maximised the data inclusion (for example mild/moderate and severe/profound). To determine the impact of severity of ID on behaviours that challenge, those with severe or profound ID were grouped together to form the target population, and levels of each behaviour that challenges within this target population were compared to the 'control', mild or moderate ID population in line with the methodology adopted by McClintock, Hall & Oliver (2003). All reports of severity of ID were included in the initial analysis, regardless of whether ID was measured by a validated measure or self-report. Living arrangement analysis compared those that were supported by individuals outside the immediate family (e.g., in a

supported group setting, residential facility or hospital) to community living arrangements (living independently or living with family or in a family-setting).

In order to determine the impact of a deficit in adaptive behaviour on behaviours that challenge, a comparison was made between those demonstrating the behaviour with a deficit and those demonstrating the behaviour without a deficit. Any reporting of adaptive behaviour was included in the analysis, regardless of whether it was from an official measure or parent self-report.

In order to determine the RR of individual syndromes, the presence of SIB, aggression and destruction among the target syndrome population was compared to the presence in either a control ID population specified by the paper (e.g. Down Syndrome, ID of mixed aetiology), or was compared to those in the sample that did not display the specific syndrome if the syndrome was not recruited specifically (e.g. the sample contained individuals with specific syndromes by chance, rather than recruiting specifically to focus on this syndrome, and therefore could be compared to the rest of the population in the sample without this syndrome). [5]

Health risk markers were calculated by comparing the number of individuals engaging in a form of behaviour that challenges with a health deficit (e.g. hearing deficit) to the number of individuals engaging in the same behaviour without said deficit.

For overactivity/impulsivity, data were included in analysis if studies reported on overactivity and/or impulsivity separately or overactivity/impulsivity combined as some measures collapse these variables.

[5] Additional studies provided data that could have been included in analysis of Down syndrome (DS), however they used DS as a control population; it was not considered appropriate to reverse compare a control population to other populations in the study, therefore these studies were not included in analysis.

1.4 Data analysis

1.4.1 Risk Markers Identified

Appendix A provides a comprehensive overview of the risk markers identified in the studies included in the meta-analysis for each of the three types of behaviour (self-injury, aggression and destruction of property). In order to generate meaningful effect sizes, it was deemed necessary for at least two papers to measure the same construct. These two papers had to have measured the construct in different sub-populations to prevent a specific phenotypic focus. Thus if repetitive behaviour had only been studied as a risk marker for aggression in two studies, it would be included if one study sampled individuals with autism, and the other study sampled individuals with tuberous sclerosis complex. However, if both studies sampled individuals with tuberous sclerosis complex, the data on repetitive behaviour as a risk marker for aggression would have been excluded from meta-analysis, as it would not be possible to determine the extent to which this represented a sub-population specific risk marker. Thus, not all of the risk markers listed in Appendix A were explored in the current meta-analysis due to insufficient available data. Table 1.5 presents the risk markers with sufficient data for exploration in the current meta-analysis.

Table 1.5 Risk markers included in meta-analysis for each behaviour subtype

	Self-Injury	Aggression	Destruction of property
Gender	√	√	√
Severity of ID	√	√	√
Adaptive behaviour deficit	√	√	*X*
Paid/congregate care	√	√	*X*
Ethnicity	*X*	√	*X*
Angelman syndrome	√	√	*X*
Tuberous sclerosis complex	√	√	*X*
Down syndrome	√	√	*X*
Fragile X syndrome	√	√	*X*
Autism	√	√	√
Smith Magenis syndrome	√	√	*X*
Cornelia de Lange syndrome	√	√	*X*
Prader Willi syndrome	√	√	*X*
Mobility deficits	√	√	*X*
Visual deficit	√	√	*X*
Skin problems	√	*X*	*X*
Gastrointestinal problems	√	*X*	*X*
Dental problems	√	*X*	*X*
Epilepsy	√	√	*X*
Hearing deficits	√	*X*	*X*
Expressive communication	√	√	*X*
Receptive communication	√	*X*	*X*
Repetitive behaviour	√	*X*	*X*
Overactivity/impulsivity	√	*X*	*X*

1.4.2 Preparation of data

Where studies presented effect size in the form of Cliffs Delta (e.g. Richards et al., 2012), this was converted using RStudio (package=orddom) to Cohen's Delta. It was subsequently possible to convert Cohen's Delta to RR using the methodology outlined in Bornstein (2009). Complexities arose for studies that presented median scores with interquartile ranges for measures (e.g. Eden et al., 2014 and Wilde et al., 2017); data were not normally distributed making it impossible to convert interquartile ranges to standard deviation. Therefore, it was not possible to calculate RR based on the presented data and thus these data were excluded from analysis. Authors were contacted for further data; however no response was received at time of writing. Relative risk is the probability of an event occurring relative to an independent

variable (Bornstein, 2009). In this meta-analysis, it would be the probability of a form of challenging behaviour occurring in a population with a target risk marker (e.g. males or autism) comparison to the control population (e.g. females or ID without autism). Relative risk is used more frequently than odds ratios in cohort or randomised control studies, and was therefore deemed most appropriate for this meta-analysis, however it is important to note that it increases to risk of overestimation of the effect in the target population (Stegenga, 2015). For the majority of data, relative risk was calculated by dividing the reported cumulative incidence in the exposed group by the cumulative incidence in the unexposed group. In such an instance, log standard error of RR could be calculated using the square root of the log variance ((1/target n)-(1/target N)+(1/control n)-(1/control N)). Where studies gave the RR rather than cumulative incidences, confidence intervals were used to calculate the log standard error of the RR by generating a mean, which was divided by square root of N. If confidence intervals were not available, and authors could not be reached, the study was removed from analysis.

1.4.3 Omnibus test

As the purpose of this meta-analysis was to synthesise data from multiple studies, methodological strengths and weaknesses between studies varied. It was therefore deemed beneficial to utilise a random effects model for analysis, as this does not assume a common effect size (as in a fixed-effects model). The goal was not to estimate one true effect but to estimate the mean of a distribution of possible effects, which may show true variation due to idiosyncratic characteristics of the individual or unique circumstances of the exposure. A random effects method adapts the weighting of each study according to its heterogeneity in comparison to the other studies included, as well as in accordance with sample size. The DerSimonian and Laird (1986) method was adopted to calculate the random effects model. This model was suitable as the effects are considered to be normally distributed within the

populations explored. Moreover, the trials are of different sizes, which would make the Sidik-Jonkman (Sidik & Jonkman, 2007) estimator less effective.

1.4.4 Forest plot and heterogeneity

Individual forest plot outputs for each behaviour and risk marker pair, calculated using RStudio package designed by Dr Chris Jones at the University of Birmingham ("Meta-Analysis of Summary Effects using Generic Inverse Variance") are presented in Appendix B for brevity. Each figure presents Te (effect) and seTE (standard error of the effect), as well as the RR, 95% confidence intervals (CI) and the percentage weight each study. RR using a random effect model is provided separately for each risk marker according to each form of behaviour (self-injury, aggression and destruction). Heterogeneity was calculated using Higgins et al. (2003) I^2 which measures the proportion of observed variance that reflects real differences in the true score. It provides a percentage score for heterogeneity. This method is recommended by Bornstein et al. (2009) for meta-analysis as it helps to indicate when subgroup or moderator analysis should be considered. Moreover, the alternative, Cochran's Q has lower power to detect heterogeneity if the number of studies included in the analysis is small and increases risk of Type 1 error. This meta-analysis includes a number of analyses involving a small number of studies, so I^2 was deemed more suitable. A small I^2 value indicates low heterogeneity and no necessity for subgroup analysis (0-50%), whilst a large value indicates high heterogeneity (75-100%) and suggests that the studies included in the meta-analysis cannot be from the same population and further analysis is required. 50-75% is considered 'moderate' (Higgins et al., 2003). To identify studies contributing most significantly to heterogeneity, the scatter plot devised by Baujat et al. (2002) was used; this presents studies in graphical form according to the extent they contribute to effect size, as well as extent they contribute to heterogeneity. This was used as the rationale for any exclusions when high heterogeneity was present; those contributing most substantively to heterogeneity were excluded one at a time to determine

impact. If heterogeneity could be reduced by 10% or greater with one or two studies, they were excluded. Where exclusion of a third study equated to a 10% or greater reduction in addition to the first one or two studies, this study was also excluded. Where heterogeneity remained high, moderator analysis was considered, for example by age, and level of intellectual disability

1.5 Results

1.5.1 Sample Characteristics

Following the procedure indicated in the methods, 51 of the 60 studies (85%) contained sufficient data to calculate RR and standard error suitable for meta-analysis.

The dates of publication of the studies ranged from 1968 to 2018. Twenty studies were conducted in the UK, seven were conducted in the USA, two in Canada, two comprised mixed country samples, two were conducted in Japan and one study came from each Italy, Jersey, Spain, Norway, Sweden, Germany and the Netherlands. The various methodologies employed were questionnaires, observations, interviews, the review of case notes/databases and the administration of published assessments.

1.5.2 Demographic Risk Markers

In order to test Part A of the aim of the meta analysis, data relating to demographic risk markers (gender, severity of ID, adaptive functioning deficit and living arrangements) was compiled for self-injury, aggression and destruction (Tables 1.6-1.8 respectively). Following this, 10 forest plots were conducted to evaluate the risk of each demographic risk marker on self-injury, aggression and destruction (Appendix B1-B4). Table 1.9 summarises the outcomes of these forest plots.

Table 1.6 Demographic and comparison risk marker data indicting number of individuals showing self-injury (n) out of the population included for each risk marker (N)

Authors	N	Sample	Quality				M:F	Gender		IQ		Adaptive functioning		Living arrangements	
								Male	Female	Mild & mod	Severe & prof	No deficit	Deficit	Paid	Family
			Q 1	Q 2	Q 3	Q 4		n	n	n/N	n/N	n/N	n/N	n/N	n/N
Arron et al. (2011)	101	CdLS					41:60	-	-	-	-	33/54	37/46	-	-
Ballinger (1971)	626	Mixed ID					343:283	46	47	25/337	68/289	-	-	-	-
Bowring et al. (2017)	265	ID					134:131	RR: 1.466	-	-	RR:5.353	-	-	RR:3/02	-
Crocker et al. (2006)	3165	Mixed ID					1633:1527	366	405	385/2169	387/996	-	-	384/1044	113/791
Crocker et al. (2007)	296	Mild/mod					162:134	10	9	-	-	-	-	16/172	1/76
Davies et al. (2016)	417	Severe ID					260:157	RR: 0.82	-	-	RR:0.78	-	-	-	-
Deb et al. (2001)	101	Mixed ID					51:50	6	18	16/90	8/11	-	-	12/37	11/48
Eden et al. (2014)	37	TSC					19:18	3	7	-	-	5/29	5/8	-	-
Eyman & Call (1977)	6870	Mixed ID					Not reported	-	-	321/4184	725/2686	-	-	-	-
Folch et al. (2008)	833	Mixed ID					432:401	59	76	137/504	53/329	-	-	78/419	57/414
Griffin et al. (1986)	11891	Mixed ID					6664:5227	761	581	-	-	-	-	-	-
Hardan & Sahl (1997)	233	Mixed ID					168:75	-	-	40/72	13/22	-	-	-	-
Hiraiwa et al. (2007)	29	PWS					14:14	-	-	-	-	-	-	-	-
Holden et al. (2002)	154	ID					89:61	-	-	10/64	43/90	-	-	-	-
Jacobson (1982)	27023	Mixed ID					14862:12160	-	-	386/12730	2315/17817	-	-	-	-
Kebbon et al. (1986)	28559	Mixed ID					Not reported	-	-	154/16746	1044/11812	-	-	-	-
Lundqvist (2013)	915	Mixed ID					504:411	152	131	196/752	82/143	-	-	-	-
Maisto et al. (1978)	1300	Mixed ID					725:575	81	101	8/306	174/994	-	-	-	-
Maurice et al. (1982)	3261	Mixed ID					1732:1529	223	180	-	-	-	-	-	-
Medeiros et al. (2013)	115	Mixed ID					80:35	20	12	-	-	-	-	-	-
Oliver et al. (2012)	970	Severe ID					589:381	-	-	-	-	-	OR3.15	-	-
Quine (1986)	399	Severe ID					245:154	32	15	-	-	-	-	-	-
Richards et al. (2012)	148	ASD					132:17	65	9	-	-	55/121	17/25	-	-
Richards et al. (2017)	207	Child ASD					181:26	RR:0.71	-	-	-	RR1.46	-	-	-
Richards et al. (2017)	216	Adult ASD					152:64	RR:0.87	-	-	-	RR:1.51	-	-	-
Rojahn (1986)	529	Mixed ID					279:250	222	209	177/233	254/293	-	-	-	-
Ross (1972)	2678	Mixed ID					Not reported	-	-	410/2485	2268/8654	-	-	-	-
Schroeder et al. (1978)	1149	Mixed ID					517:632	109	99	14/155	194/995	-	-	-	-
Wilde et al. (2017)	29	TSC					18:9	6	3	-	-	1/9	10/20	-	-

Table 1.7 Demographic and comparison risk marker data indicting number of individuals showing aggression (n) out of the population included for each risk marker (N)

Author	N	Sample	Quality				M:F	Gender		IQ		Adaptive function	
			Q 1	Q 2	Q 3	Q 4		Male n	Female n	Mild & mod n/N	Severe & prof n/N	No deficit n/N	Deficit n/N
Arron et al. (2011)	189	PWS					100:89	64	32	-	-	15/49	25/52
Bowring et al. (2017)	265	ID					134:131	RR:1.173	-	-	RR:2.471	-	-
Cohen et al. (2009)	3547	Mixed ID					Not reported	-	-	357/1422	529/2060	-	-
Cooper et al. (2009)	1023	ID					562:461	42	58	43/646	57/377	-	-
Crocker et al. (2006)	3165	ID					1633:1527	839	799	371/1527	397/1633	-	-
Crocker et al. (2007)	296	Mild/mod ID					162:134	31	24	-	-	-	-
Davidson et al. (1994)	201	Mixed ID					126:79	89	42	82/129	49/70	-	-
Davies et al. (2016)	417	Severe ID					258:159	RR: 0.74	-	RR:1.19	-	-	-
Deb et al. (2001)	101	Mixed ID					51:50	8	15	18/80	5/11	-	-
Eden et al. (2014)	37	TSC					19:18	14	4	10/13	7/19	3/8	15/29
Medeiros et al. (2014)	115	Mixed ID					80:35	41	17	-	-	-	-
Eyman & Call (1977)	6870	Mixed ID					Not reported	-	-	1229/4381	899/2489	-	-
Hardan & Sahl (1997)	233	Mixed ID					168:75	-	-	14/72	13/22	-	-
Jacobson (1982)	30577	Mixed ID					14862:12160	-	-	899/12730	2651/17847	-	-
Quine (1986)	399	Mixed ID					245:154	56	26	-	82/399	-	-
Ross (1972)	11139	Mixed ID					Not reported	-	-	731/2485	1885/8654	-	-
Tenneij et al. (2009)	108	Mild LD					40:48	16	23	-	-	-	-
Tyrer et al. (2005)	3065	Mixed ID					1735:1327	287	156	127/1317	299/1647	-	-
Wilde et al. (2017)	29	TSC					18:9	6	5	-	-	7/18	4/11

Table 1.8 All risk marker data (Gender, IQ and Autism) indicting number of individuals showing destruction (n) out of the population included for each risk marker (N)

Authors	N	Sample	Quality				M:F	Gender		IQ		ASD	
			Q 1	Q 2	Q 3	Q 4		Male n	Female n	Mild & mod n/N	Severe & prof n/N	ASD n/N	Control n/N
Ando et al. (1979)	175	Mixed ID					Not reported	-	-	-	-	16/47	5/128
Bhaumick et al. (1997)	2201	Mixed ID					Not reported	-	-	-	-	263/1044	344/1157
Crocker et al. (2006)	3165	Mixed ID					1633:1527	-	-	498/2169	283/996	-	-
Crocker et al. (2007)	296	Mixed ID					162:134	105	80	-	-	-	-
Davies et al. (2016)	417	Severe ID					258:159	RR: 0.81	-	-	RR: 0.84	-	-
Jacobson (1982)	32568	Mixed ID					Not reported	-	-	1456/13633	3128/18935	-	-
Matson et al. (2008)	320	ASD/ID					179:141	-	-	-	-	15/62	15/159
Sheth et al. (2015)	150	ASD/DS					101:49	-	-	-	-	20/38	4/38

Table 1.9 Synthesis of risk markers relating to Aim 1 (impact of demographic risk markers on three forms of behaviour that challenges) and results of prior relative risk analysis in McClintock study

Risk Marker	**Behaviour**	**Number of studies included**	**McClintock et al. (2003) RR**	**RR** ***(95% CI)***	**Z score**	***p***	**Heterogeneity (I2, %)**	**Excluded studies**
Gender	Self-injury	17	0.97	0.93 *(0.87 – 0.99)*	-2.15	.031*	20.2	Maisto et al. (1978); Schroeder et al. (1978); Deb et al. (2001)
	Aggression	11	1.71*	1.04 *(0.86 - 1.25)*	0.36	.717	80.8	-
	Destruction	3	Not assessed	1.07 *(0.90 - 1.26)*	0.73	.464	0.0	-
IQ	Self-injury	16	4.06**	2.39 *(1.67 – 3.41)*	4.90	<.001**	98.0	-
	Aggression	9	1.28	1.24 *(1.04 – 1.48)*	2.43	.015*	84.0	Jacobson et al. (1982), Ross et al. (1972), Tyrer et al. (2006)
	Destruction	3	Not assessed	1.30 *(1.03 - 1.64)*	2.22	.026*	88.0	-
Adaptive behaviour	Self-injury	6	Not assessed	1.48 *(1.20 – 1.82)*	3.64	<.001**	9.2	-
	Aggression	3	Not assessed	0.71 *(0.47 – 1.08)*	1.61	.110	0.0	-
Living arrangements	Self-injury	4	Not assessed	2.36 *(1.63 – 3.42)*	4.56	<.001**	17.8	Folch et al. (2018)
	Aggression	5	Not assessed	1.86 *(1.60 – 2.16)*	8.16	<.001**	22.9	-

*Sig at p<.05 level; **Sig at p<.001 level;

1.5.2.1 Gender

SIB was the only form of behaviour that challenges to be associated with gender with the RR of SIB being 9% lower among males than females (RR=0.93, CI=0.87, 0.99; p<.05). Moderator analysis by age accounted for 16.13% of residual heterogeneity. There was no significant impact of gender on aggression or destruction. Given that only three studies were included to assess the impact of gender on destructive behaviour, these results should be interpreted with caution.

1.5.2.2. Severity of ID

Individuals with severe and profound levels of ID had 143% greater risk of self-injury than those with mild or moderate levels (RR=2.39, CI=1.67, 3.41; *p*<.001). A 24% increased risk was also identified for aggression (RR=1.24, CI=1.04, 1.48; *p*<.05). Heterogeneity could not be accounted for using study exclusion or using moderator variable of age for self-injury. There was a 30% increased risk of destruction among those with severe/profound ID (RR=1.30, CI=1.03, 1.64; *p*<.05), however as only based on three studies with high heterogeneity, the results should be interpreted with caution.

1.5.2.3 Adaptive behaviour deficit

Individuals with a deficit in adaptive behaviour had 48% greater risk of self-injury (RR=1.48, CI=1.20, 1.82; *p*<.001). There was no significant relationship between adaptive function and aggression, however this is only based on three studies. Insufficient data were available to assess the impact of adaptive behaviour on destruction.

1.5.2.4 Living Arrangements (congregate/paid care)

There was a 136% increased risk of self-injury among those living in paid/congregate care compared to those living with family (RR=2.36, CI=1.63, 3.42, *p* <.001) and an 86% increased

risk of aggressive behaviour (RR=1.86, CI=1.60, 2.16; p <.001), however this was only based on five studies with high heterogeneity, and therefore should be interpreted with caution. Insufficient data were available to assess the impact of paid/congregate care on destructive behaviour.

1.5.3 Given diagnosis risk markers

In order to address Part B of the aim of the meta-analysis, data relating to specific syndromes or diagnoses (Angelman syndrome (AS), Autism (ASD), tuberous sclerosis complex (TSC), Down syndrome (DS), fragile X syndrome (FXS), Autism, Smith Magenis syndrome (SMS), Cornelia de Lange syndrome (CdLS), and Prader Willi syndrome (PWS)) were compiled for self-injury, aggression and destruction (Tables 1.10, 1.11 & 1.8[6]). 17 forest plots (Appendix B5-B12) were conducted to determine the RR of different syndromes on each of the three behaviours. These are summarised in Table 1.12.

[6] All data pertaining to destruction is comprised in Table 8 due to so little being available.

Table 1.10 Syndrome and control (con) risk marker data indicting number of individuals showing self-injury (n) out of the population included for each risk marker (N)

Author	N	Sample	Quality				AS		TSC		DS		FXS		ASD		SMS		CdLS		PWS	
			Q 1	Q 2	Q 3	Q 4	Agm n/ N	Con n/ N	DS n N	Con n/ N	TSC n/ N	Con n/ N	FXS n/ N	Con n/ N	ASD n/ N	Con n/ N	SMS n/ N	Con n/ N	CdL n/ N	Con n/ N	PWS n/ N	Con n/ N
Ando et al. (1979)	175	Mixed ID/ASD[2]					-	-	-	-	-	-	-	-	20/ 47	7/ 128	-	-	-	-	-	-
Arron et al. (2011)	892	CDLS/SMS/ AS/FXS[1]					47/ 104	13/ 47	-	-	-	-	98/ 191	40/ 151	-	-	88/9 5	40/ 151	71/ 101	40/ 151	294/ 571	40/ 151
Bowring et al. (2017)	265	Mixed ID[2]					-	-	-	-	RR 0.34	-	-	-	RR 1.20 8							
Bhaumik et al. (1997)	2201	Mixed ID[2]					-	-	-	-	-	-	-	-	282/ 1044	101/ 1157	-	-	-	-	-	-
Eden et al. (2014)	441	TSC/DS/FXS/ ASD[1]					-	-	13/ 37	5/ 43	-	-	61/ 112	5/ 43	78/ 188	5/ 43	-	-	39/ 61	5/ 43	-	-
Emerson et al. (2001)	95	Mixed ID[2]					-	-	-	-	-	-	-	-	RR 1.7	-	-	-	-	-	-	-
Folch et al. (2008)	833	IDD[2]					-	-	-	-	-	-	-	-	59/ 158	77/ 675	-	-	-	-	-	-
Hiraiwa et al. (2007)	178	PWS[2]					-	-	-	-	-	-	-	-	-	-	-	-	-	-	17/ 29	2/ 42
Langthorne et al. (2012)	83	FXS/SMS[2]					-	-	-	-	-	-	19/ 34	18/ 30	-	-	21/ 25	18/ 30	-	-	-	-
Lundqvist (2013)	915	ID[2]					-	-	-	-	4/ 14	279/ 901	4/ 14	279/ 901	72/ 143	211/ 772	-	-	-	-	5/9	278/ 816
Matson et al. (2008)	320	ASD/ID[2]					-	-	-	-	-	-	-	-	53/ 161	15/ 159	-	-	-	-	-	-
Oliver et al. (2009)	100	CdLS/ID[2]					-	-	-	-	-	-	-	-	-	-	-	-	30/5 4	19/4 6	-	-
Richards et al. (2012)	321	ASD/FXS[1]					-	-	-	-	-	-	67/ 123	9/ 49	74/ 148	9/ 49	-	-	-	-	-	-
Sheth et al. (2015)	150	ASD/PWS/FX[1]					-	-	-	-	-	-	-	-	19/ 38	3/ 38	-	-	-	-	26/ 38	3/ 38
Wilde et al. (2017)	50	TSC/AS[1]					13/ 29	4/ 21	9/ 29	4/ 21	-	-	-	-	-	-	-	-	-	-	-	-

[1] *Contrast group: Down syndrome*

[2] *Contrast group: Mixed ID*

Table 1.11 Syndrome and control (con) risk marker data indicting number of individuals showing aggression (n) out of the population included for each risk marker (N)

Author	N	Sample	Quality				AS		DS		TSC		FXS		ASD		SMS		CdLS		PWS	
			Q 1	Q 2	Q 3	Q 4	AS n/N	Con n/N	DS n/N	Con n/N	TSC n/N	Con n/N	FXS n/N	Con n/N	ASD n/N	Con n/N	SMS n/N	Con n/N	CdLS n/ N	Con n/N	PWS n/N	Con n/N
Ando et al. (1979)	175	Mixed ID/ASD[2]					-	-	-	-	-	-	-	-	12/ 47	6/ 128	-	-	-	-	-	-
Arron et al. (2011)	892	CDLS/SMS, AS/FXS[1]					76/ 104	22/ 47	-	-	-	-	100/ 191	70/ 151	-	-	70/ 95	70/ 151	57/ 142	70/ 151	97/ 189	40/ 151
Bhaumik et al. (1997)	2201	Mixed ID[2]					-	-	-	-	-	-	-	-	318/ 1044	159/ 1157	-	-	-	-	-	-
Cooper et al. (2009)	1023	Mixed ID[2]					-	-	4/ 186	96/ 837	-	-	-	-	9/ 77	91/ 946	-	-	-	-	-	-
Davidson et al. (1994)	201	Mixed ID[2]					-	-	-	-	-	-	-	-	7/10	124/ 189	-	-	-	-	-	-
Eden et al. (2014)	441	TSC/DS/FXS/ ASD[1]					-	-	-	-	18/ 37	17/ 43	68/ 112	17/ 43	125/ 188	17/43	-	-	27/ 61	18/ 43	-	-
Hiraiwa et al. (2007)	178	Mixed ID[2]					-	-	-	-	-	-	-	-	-	-	-	-	-	-	17/ 29	2/42
Langthorne et al. (2011)	83	FXS/SMS[2]					-	-	-	-	-	-	18/ 34	20/ 30	-	-	21/25	19/ 30	-	-	-	-
Lundqvist et al. (2013)	915	Mixed ID[2]					-	-	-	-	-	-	-	-	-	-	-	-	-	-	5/9	278/ 906
Matson et al. (2008)	320	ASD/ID[2]					-	-	-	-	-	-	-	-	17/ 62	23/ 159	-	-	-	-	-	-
Oliver et al. (2009)	100	CdLS/ID[2]					-	-	-	-	-	-	-	-	-	-	-	-	17/ 54	22/ 46	-	-
Sheth et al. (2015)	150	ASD/PWS/FXS/ Sotos[1]					-	-	-	-	-	-	-	-	21/36	11/ 38	-	-	-	-	26/ 28	11/ 38
Tyrer et al. (2005)	3065	Mixed ID[2]					-	-	30/ 502	413/ 2760	-	-	-	-	20/ 68	423/ 2994	-	-	-	-	-	-
Wilde et al. (2017)	50	TSC/AS[1]					21/ 29	4/ 29	-	-	11/ 29	4/ 21	-	-	-	-	-	-	-	-	-	-

[1] *Contrast group: Down syndrome*

[2] *Contrast group: Mixed ID*

Table 1.12 Synthesis of risk markers relating to Aim 2 (impact of diagnosis on three forms of behaviours that challenge)

Risk Marker	Behaviour	Number of studies	McClintock et al. (2003) RR	RR *(95% CI)*	*Z* score	Significance	Heterogeneity (I^2, %)	Excluded studies
Angelman syndrome	Self-injury	2	Not assessed	1.77 *(1.13 - 2.78)*	3.52	<.001**	0.0	-
	Aggression	2	Not assessed	2.20 *(0.95 – 5.50)*	1.85	.064	71.1	-
Autism	Self-injury	8	6.41**	2.86 *(2.18 – 3.75)*	7.56	<.001**	67.0	Ando et al. (1979)
	Aggression	6	2.79*	2.04 *(1.79 – 2.34)*	20.37	<.001**	0.0	Davidson et al. (1994)
	Destruction	3	5.60**	2.58 *(2.14 – 3.10)*	10.06	<.001**	0.0	Ando et al. (1979)
Cornelia de Lange syndrome	Self-injury	3	Not assessed	2.48 *(1.31 – 4.71)*	2.78	.005*	85.0	-
	Aggression	3	Not assessed	0.87 *(0.68 – 1.07)*	-1.09	0.281	14.6	-
Down syndrome	Self-injury	3	Not assessed	0.46 *(0.33 – 0.63)*	-4.65	<.001**	0.0	-
	Aggression	2	Not assessed	0.32 *(0.16 – 0.62)*	-3.33	<.001**	49.8	-
Fragile X syndrome	Self-injury	3	Not assessed	2.66 *(1.63 – 4.34)*	-3.93	<.001**	57.4	Langthorne et al. (2012); Lundqvist (2013)
	Aggression	3	Not assessed	1.12 *(0.82 – 1.53)*	0.71	0.475	61.4	-
Prader Willi syndrome	Self-injury	4	Not assessed	1.91 *(1.47 – 2.48)*	4.82	<.001**	0.0	Hiraiwa et al. (2007)
	Aggression	2	Not assessed	1.23 *(0.81 – 1.26)*	0.61	0.540	72.4	-
Smith Magenis syndrome	Self-injury	2	Not assessed	2.14 *(0.91 – 5.01)*	1.74	0.083	88.6	-
	Aggression	2	Not assessed	1.43 *(1.13 – 1.81)*	3.01	.003*	0.0	-
Tuberous sclerosis complex	Self-injury	2	Not assessed	2.29 *(1.14 – 4.61)*	2.33	.020*	0.0	-
	Aggression	2	Not assessed	1.35 *(0.87 – 2.10)*	1.35	.181	0.0	-

1.5.3.1 Angelman syndrome

There was a 76% risk of self-injury among individuals diagnosed with AS compared to those of mixed aetiology ID *(*RR= 1.77, CI = 1.13, 2.78; p = <.001). Whilst the RR of aggression among those with AS approached significance, it was not significant based on the two studies analysed. Insufficient information was available to assess destruction.

1.5.3.2 Autism

There was a 186% increased risk of self-injury among individuals with autism in addition to ID, compared to those with ID alone (RR=2.86, CI=2.18, 3.75; p<.001). Similar results were found for aggression, with individuals diagnosed with autism having 104% increased risk of aggressive behaviour (RR=2.04, CI=1.79, 2.34; p<.001). A significant impact of autism was also found for destructive behaviour, with a 158% increased risk (RR=2.58, CI=2.14, 3.10; p<.001).

1.5.3.3 Cornelia de Lange syndrome

The analysis indicated that those with CdLS have a 148% increased risk of self-injury *(RR*=2.48, CI=1.31, 4.71; p =.005). There was no significant effect of CdLS on aggressive behaviour, however both analyses were based on a small number of studies and should be interpreted with caution. Insufficient information was available to assess destruction.

1.5.3.4 Down syndrome

Based on the three studies available, individuals with DS have a 54% reduction in risk of self-injury compared to other forms of ID (RR=0.46, CI=0.33, 0.63; p<.001). A similar result was found for risk of aggression; individuals with DS has a 68% reduced risk (RR=0.32, CI=0.16, 0.62; p<.001). Insufficient information was available to assess destruction.

1.5.3.5 Fragile X syndrome

The analysis indicated that individuals with FXS have a 166% increased risk of self-injury above those with ID of other aetiology (RR=2.66, CI=1.63, 4.64, $p<.001$). Based on the three studies focussing on aggression, there was no significant impact of FXS on aggressive behaviour. Insufficient information was available to assess destruction.

1.5.3.6 Prader Willi syndrome

Based on the two studies included in the analysis, individuals with PWS have a 91% increased risk of self-injury than other ID aetiologies (R =1.91, CI=1.47, 2.48; $p<.001$). No significant effect of PWS was found on aggressive behaviour. Insufficient information was available to assess destruction.

1.5.3.7 Smith Magenis syndrome

Based on the two available studies, individuals with SMS were at no increased risk for self-injury than individuals of mixed aetiology ID. The results however indicate a 43% increased risk of aggression among individuals with SMS (RR=1.43, CI=1.13, 1.81; $p = .003$). Insufficient information was available to assess destruction.

1.5.3.8 Tuberous sclerosis complex

Based on the two studies available, individuals with TSC had a 129% greater risk of self-injury than individuals of mixed aetiology ID (RR=2.29, CI=1.14, 4.61; $p = .02$). There was no significant impact of TSC on aggression and insufficient information available for destruction.

1.5.4 Health Risk Markers

In order to address Part C of the meta-analysis aim, data relating to specific health risk markers (mobility, vision, hearing, skin, gastrointestinal, dental and epilepsy) were compiled for self-injury and aggression (Tables 1.13 & 1.14). These data were not available for destruction. 7

forest plots (Appendix B13-B19) were conducted to determine the RR of different health markers on each of the behaviours. These are summarised in Table 1.15.

Table 1.13 Deficit and typical health marker data indicting number of individuals showing self-injury (n) out of the population (N)

Author	N	Sample	Quality				Mobility		Vision		Skin		Gastro		Dental		Epilepsy		Hearing	
			Q 1	Q 2	Q 3	Q 4	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/ N
Bowring et al. (2017)	265	Mixed ID					RR 1.89	-	RR 2.285	-	-	-	-	-	-	-	RR 2.971		RR 0.505	-
Deb et al. (2001)	101	Mixed ID					-	-	-	-	-	-	-	-	-	-	8/25	15/76	-	-
Eden et al. (2014)	37	TSC					2/7	8/30	2/4	8/33	-	-	-	-	-	-	-	-	-	-
Folch et al. (2008)	833	Mixed ID					-	-	-	-	-	-	-	-	10/18	85/189	41/242	94/591	-	-
Kiernan et al. (1996)	34	Mixed ID					7/9	13/25	-	-	-	-	-	-	-	-	-	-	-	-
Lundqvist (2013)	915	Mixed ID					-	-	12/35	270/880	-	-	-	-	-	-	56/124	264/874	5/6	271/881
Richards et al. (2012)	148	ASD					9/13	64/134	5/6	69/141	-	-	-	-	-	-	-	-	5/6	69/142
Richards et al. (2017)	207	ASD					-	-	3/7	92/200	24/35	71/172	18/36	77/104	10/18	85/189	-	-	-	-
Schroeder et al. (1978)	200	Mixed ID					-	-	45/52	55/148	-	-	-	-	-	-	-	-	16/21	84/179
Wilde et al. (2017)	29	TSC					4/10	5/19	1/6	8/23	4/15	5/14	4/5	5/24	1/3	8/26	8/23	1/6	-	-

Table 1.14 Deficit and typical health marker data indicting number of individuals showing aggression (n) out of the population (N)

Author	N	Sample	Quality				Vision		Epilepsy		Mobility		Expressive Communication	
			Q 1	Q 2	Q 3	Q 4	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N	Deficit n/N	Typical n/N
Cooper et al. (2009)	1023	Mixed ID					55/481	45/542	42/349	56/663	22/248	78/775	56/124	264/874
Deb et al. (2001)	101	Mixed ID					-	-	9/25	14/76	-	-	9/25	14/76
Eden et al. (2014)	37	TSC					2/3	17/34	-	-	1/9	1/16	-	-
Tyrer et al. (2005)	2062	Mixed ID					-	-	152/812	291/2250	-	-	-	-
Wilde et al. (2017)	29	TSC					-	-	-	-	4/10	6/19	5/15	4/14

Table 1.15 Synthesis of risk markers relating to Aim 3 (impact of health risk markers on all forms of behaviour that challenges)

Risk Marker	**Behaviour**	**Number of studies included**	**McClintock et al. (2003) RR**	**RR** *(95% CI)*	***Z* score**	**Significance**	**Heterogeneity** (I^2)	**Excluded studies**
Visual deficit	Self-injury	5	Not assessed	2.09 *(1.67 – 2.60)*	6.53	<.001**	5%	Lundqvist (2013)
	Aggression	2	Not assessed	1.37 *(0.97-1.93)*	1.81	.07	0%	-
Hearing deficit	Self-injury	3	Not assessed	1.54 *(1.24 – 1.90)*	4.0	<.001**	0%	Bowring et al. (2017)
Mobility deficit	Self-injury	5	Not assessed	1.46 *(1.08 – 1.96)*	2.49	.013*	0%	-
	Aggression	3	Not assessed	0.95 *(0.63 – 1.42)*	-0.25	0.812	0%	-
Epilepsy	Self-injury	4	Not assessed	1.37 *(1.15 – 1.64)*	3.58	<.001**	0%	-
	Aggression	3	Not assessed	1.46 *(1.25 – 1.71)*	4.80	<.001**	0%	-
Dental Problems	Self-injury	3	Not assessed	0.94 *(0.75 – 1.19)*	-.48	.631	0%	-
Gastro problems	Self-injury	2	Not assessed	1.39 *(0.31 – 6.33)*	.43	.673	0%	-
Skin problems	Self-injury	3	Not assessed	1.34 *(0.67 – 2.69)*	.82	.412	0%	-

*Sig at p<.05 level; **Sig at p<.001 level;

1.5.4.1 Visual deficit

The analysis indicated that people with visual deficits and ID have a 109% increased risk of self-injury compared to those with ID and no deficit (RR =2.09, CI=1.67, 2.60; p<.001). The impact of visual deficit on aggressive behaviour approached significance but was not significant based on the two studies available. Insufficient data were available to determine impact on destructive behaviour.

1.5.4.2 Hearing Deficit

The analysis indicated that individuals with ID with a deficit in hearing have a 54% increase in risk of self-injury compared to those without this deficit (RR=1.45, CI=0.61-1.91; p = .006). Insufficient data were available to determine impact on aggression and destructive behaviour.

1.5.4.3 Mobility Deficit

The forest plots indicated that individuals with ID and a mobility deficit are at a 46% increased risk of self-injury (RR=1.46, CI=1.08, 1.96; p = .013). No significant effect of deficit in mobility was found on risk of aggression. Insufficient data were available to determine impact on aggression and destructive behaviour.

1.5.4.4 Epilepsy

The analysis indicated that individuals with epilepsy and ID have 64% greater risk of self-injury compared to those with ID alone (RR=1.37, CI=1.15, 1.64; p = <.001). A similar result was identified for aggression; individuals with epilepsy and ID have a 46% increased risk of aggression compared to those with ID alone (RR=1.46, CI=1.25, 1.71; p<.001). Insufficient data were available to determine impact on destructive behaviour.

1.5.5 Person Risk Markers

To further address the Part C of the meta-analysis aim, data relating to specific person risk markers (receptive and expressive communication, repetitive behaviour and

overactivity/impulsivity) were compiled for self-injury and aggression (Tables 1.14[7] & 1.16). These data were not available for destruction in the literature. 5 forest plots (Appendix B20-B23) were conducted to determine the RR of different person risk markers on self-injury. These are summarised in Table 1.17.

[7] Only one person risk marker was available for aggression (expressive communication), therefore data is included in previous aggression table, Table 1.14.

Table 1.16 Person specific risk marker data indicting number of individuals showing self-injury (n) out of the population (N)

Author	N	Sample	Quality				Expressive communication		Receptive communication		Repetitive behaviour	Overactivity/ impulsivity
							Deficit	Typical	Deficit	Typical	Deficit	High score
			Q 1	Q 2	Q 3	Q 4	n/N	n/N	n/N	n/N	RR	RR
Ando et al. (1979)	128	Mixed ID					3/99	4/29	5/118	2/11	-	-
Bott et al. (1997)	3662	LD					383/2994	173/668	-	-	-	-
Bowring et al. (2017)	265	ID					-	RR: 3.68	-	RR: 3.658	-	-
Davies et al. (2016)	417	Severe ID					-	-	-	-	RR: 4.87	RR: 3.51
Eden et al. (2014)	37	TSC					8/33	2/4	-	-	-	
Emerson et al. (2001)	95	Mixed ID					RR: 2.2		RR:1.7	-	-	-
Hemmings et al. (2006)	214	ID					-	-	-	-	-	-
Kiernan et al. (1996)	42	ID					-	-	7/24	8/18	-	-
Lundqvist (2013)	915	ID					234/769	78/146	-	-	-	-
McLean et al. (1996)	211	Mixed ID					23/166	6/45	-	-	-	-
Richards et al. (2012)	148	ASD					60/130	14/18	-	-	-	*RR1.758 *RR1.614
Richards et al. (2017)	148	ASD					-	-	-	-	*RR: 1.41 *RR: 2.5	*RR: 2.23 *RR: 2.12
Schroeder et al. (1978)	1153	Mixed ID					62/819	146/334	136/956	72/194	-	-
et al. (1968)	59	ID/Schizophrenia					4/21	18/38	-	-		
Wilde et al. (2017)	29	TSC					3/14	6/15	-	-	-	-

*Two results given as considered overactivity and impulsivity separately.

Table 1.17 Synthesis of risk markers relating to Aim 3 (impact of person risk markers on self-injury and aggression)

Risk Marker	**Behaviour**	**Number studies included**	**McClintock et al. (2003) RR**	**RR** ***(95% CI)***	***Z* score**	**Significance**	**Heterogeneity** **(I^2)**	**Excluded studies**
Expressive communication	Self-injury	8	3.37**	1.95 *(1.70 – 2.23)*	9.74	<.001**	0%	Schroeder et al. (1978); Lundqvist (2013)
	Aggression	5	0.46	1.44 *(1.29 – 1.60)*	6.46	<.001**	0%	McLean et al. (1996)
Receptive communication	Self-injury	4	3.43**	2.54 *(2.03 – 3.17)*	8.21	<.001**	0%	-
Overactivity/impulsivity	Self-injury	5	Not assessed	1.65 (*1.49 – 1.84)*	9.37	<.001**	0%	-
Repetitive behaviour	Self-injury	3	Not assessed	1.66 *(0.97 – 2.84)*	1.85	.065	0%	-

*Sig at p<.05 level; **Sig at p<.001 level

1.5.5.1 Expressive Communication Deficit

The results indicate a 95% increase in risk of self-injury for individuals with a deficit in expressive communication compared to those without (RR=1.95, CI=1.7, 2.23; p = <.001). A similar result was identified for aggression; individuals with an expressive communication deficit have a 44% increased risk of aggressive behaviour compared to those without (RR=1.44, CI=1.29, 1.60*;* p<.001).

1.5.5.2 Receptive Communication Deficit

The analysis concluded that individuals with a deficit in receptive communication have 154% increased risk of self-injury than those without this deficit (RR = 2.54, CI = 2.03, 3.17; p <.001). Insufficient data were available to determine impact on aggression and destructive behaviour.

1.5.5.3 Overactivity/Impulsivity

The forest plot indicates that individuals who score more highly on measures as being overactive or impulsive are 65% more likely than those who do not to self-injure (RR=1.65, CI=1.49, 1.84; p<.001). Insufficient data were available to determine impact on aggression and destructive behaviour.

1.5.5.4 Repetitive Behaviour

Based on the two studies available for analysis, the impact of repetitive behaviour on self-injury approached significance, however it not currently possible to conclude that repetitive behaviour impacts the risk of self-injury. Insufficient data were available to determine impact on aggression and destructive behaviour.

1.5.6 Summary of significant results

Figure 1.2 provides a synthesis of all statistically significant risk markers identified throughout the meta-analysis.

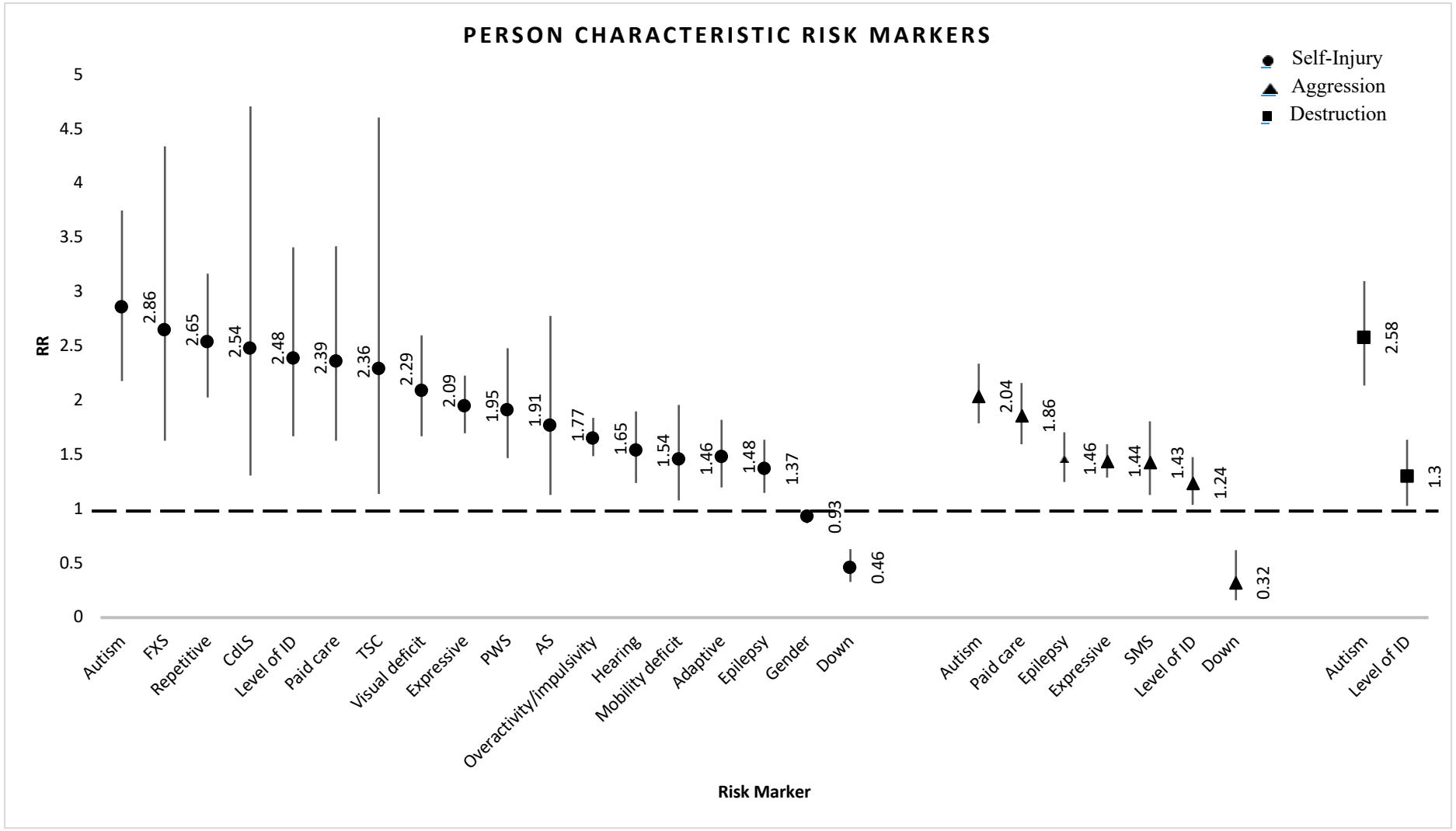

Figure 1.2 Synthesis of all significant risk markers for self-injury, aggression and destruction, showing relative risk and associated confidence intervals.

1.6. Discussion

The current study sought to synthesise demographic, diagnostic and behavioural risk markers for three distinct forms of behaviour; SIB, aggression and destruction. It provided a synthesis of risk marker research from across 60 studies conducted between 1968 and 2018. It has highlighted considerable progression in the understanding of risk markers associated with SIB and has indicated that aggressive and destructive behaviour risk markers warrant considerable further research to meet the same level of understanding as risk markers for SIB. Moreover, this meta-analysis has indicated methodological concerns associated with conflating aggressive and destructive behaviour, as it results in lack of understanding about each specific topography.

1.6.1 Self-injury

This meta-analysis identified 18 risk markers that were significantly associated with SIB. A diagnosis of autism was associated with the greatest increase in risk of SIB (RR=2.86), along with a diagnosis of fragile X syndrome (RR=2.65), increased repetitive behaviour (RR=2.54) diagnosis of Cornelia de Lange syndrome (RR=2.48), lower level of intellectual ability (RR=2.39), living in paid care (RR=2.36), diagnosis of tuberous sclerosis complex (RR=2.29) and a visual deficit (RR=2.09). All of these risk markers are associated with over a 100% increase in risk of SIB when present, compared to individuals with ID that do not have these characteristics. This meta-analysis also identified expressive communication deficit (RR=1.95), diagnosis of Prader Willi syndrome (RR=1.91), Angelman syndrome (RR=1.77), overactivity (RR=1.65), hearing deficit (RR=1.54), mobility deficits (RR=1.46), low adaptive functioning (RR=1.48) and epilepsy (RR=1.37) as risk markers for SIB. Finally, male gender was associated with a decrease in risk of SIB (RR=0.93), as was diagnosis of Down syndrome (RR=0.46).

The previous meta-analysis identified diagnosis of autism as a risk marker for SIB, as well as severity of ID and deficits in receptive and expressive communication (McClintock et al., 2003). The reliability of these risk markers has been upheld and refined with the addition of further research that has emerged, suggesting that these are robust and reliable risk markers. Many of the additional risk markers in the new analysis, however, represent emerging interests within the field of intellectual disability and SIB. In the same manner as the previous meta-analysis, many of the new risk markers identified are based on a small number of studies, such as Angelman syndrome (N=2), Cornelia de Lange syndrome (N=3), Down syndrome (N=3), fragile X syndrome (N=3), Smith Magenis syndrome (N=2) and Tuberous sclerosis complex (N=2). The identification of these new risk markers provide guidance for future research focus, to enhance the robustness of these findings.

1.6.2 Aggression

The results of the meta-analysis of risk markers pertaining to aggression suggest that the research field has focused on similar risk markers to SIB. Importantly, however, many of the risk markers that are significantly associated with SIB have not been replicated for aggression. Seven risk markers were identified in this meta-analysis: diagnosis of autism (RR=2.04), living in paid care (RR=1.86), epilepsy (RR=1.46), deficit in expressive communication (RR=1.44), diagnosis of Smith Magenis syndrome (RR=1.43), severity of ID (RR=1.24) and Down syndrome (RR=0.32). Three of these were consistent with the McClintock et al. (2003) paper (autism, expressive communication and severity of ID), however gender was previously indicated as a significant risk marker that was not upheld in the current study. This was, however, previously based on two studies and is now based on eleven; it is likely the additional studies helped to increase the robustness of this finding and emphasises the importance of contributing additional studies to the field of aggression risk markers to help refine current understanding. Research pertaining to specific syndromes and their association with aggression

suggests a similar situation to SIB; the number of studies available is relatively small. There is considerable merit in more widespread research within these less well-studied syndromes, as well as consideration of more diverse risk markers that may be unique to aggression, rather than focusing solely on those identified in SIB research.

1.6.3 Destruction

This meta-analysis has emphasised the noticeable lack of research focussing on risk markers for destruction compared to both aggression and SIB. It was only possible to identify eight papers that provide sufficient data for analysis on destruction among individuals with ID, and these papers only explored three risk markers, of which two were significant: severity of ID (RR=2.58) and presence of autism (RR=2.58). There is considerable diversity in the proportion of individuals displaying destruction in the samples identified, with the highest being 48.5% (Crocker et al., 2007), a mid-range proportion being 24.7% (Crocker et al., 2006) and the smallest proportion being 14.1% (Jacobson et al., 1982). This suggests that there may be some inconsistencies in the definition of destruction and emphasises the importance of consistency in better understanding different topographies of behaviour.

Across each of the three behaviours studies, the presence of autism and lower level of ability have been identified as significant risk markers. Interestingly, a recent meta-analysis indicated that individuals with autism and ID are often excluded from research, when this is arguably the population most in need (Russel et al., 2019). There are considerable clinical implications of being able to identify individuals that are at increased risk of developing either of these three behaviours. It is widely recognised that, once these behaviours are in a person's repertoire, it is difficult to exclude them, resulting in incredibly high costs to the NHS (cost to services for adults with severe challenging behaviour was indicated as being between £89,335 and £358,415; Emerson et al., 2014). Future research should 1) consider how to use these risk

markers to identify individuals most at risk in a clinical setting and 2) devise intervention or prevention strategies tailored towards those with increased risk.

1.6.4 Research progression

The differences identified between the previous meta-analysis and the current meta-analysis provide a clear indication of the value of having multiple studies to assess. Inclusion of a small number of studies increases the risk of both Type 1 and Type 2 errors. This meta-analysis provides an opportunity to appraise the updated literature base (Blettner et al., 1999), considering the impact the additional research has made to the previous findings presented by McClintock, Hall and Oliver (2003). In comparison to the 25.5% inclusion rate McClintock, Hall and Oliver (2003) indicate for identified studies containing sufficient data to calculate effect size, the current study had an 85% inclusion rate of identified studies. This suggests considerable improvement in the way that data are reported. There has been increased consideration for the inclusion of control groups and more comprehensive information regarding the ID population included in research, two factors highlighted by McClintock et al. (2003) as deficits in the literature base. This increased detail facilitates more reliable reporting on unique characteristics, rather than the general ID population. Despite this improvement, the analysis indicates that this is predominantly within self-injury; less researched fields of aggression and destruction of property still require advancements to enable direct comparisons, particularly in terms of measures used.

1.6.5 Limitations

A potential limitation of the current meta-analysis is that it elected not to rerun the search that was previously conducted by McClintock et al. (2003). It is possible, that choosing to extend the search opens up the possibility of missing valuable studies that the previous meta-analysis may have missed or excluded. Given that the search terms included in the current meta-

analysis, however, were based on the previously used search terms, it is unlikely that a new search would have identified any additional papers. Moreover, the bibliographies of the included studies were reviewed for any additional studies that may have been overlooked. It is anticipated that this thorough approach would have identified any studies not identified in the database search. This decision was conducted in line with guidance on updating previous comprehensive reviews (Garner et al., 2016). Similarly, the methodology could be criticised for limiting the search solely to peer reviewed, published studies, as this increases the risk of positive publication bias influencing the results. However it was considered of greater value to include studies that have been peer-reviewed as these represent a higher standard (Cook et al., 2003).

In line with the previous meta-analysis (McClintock et al., 2003), there was considerable heterogeneity (indicated by high I^2 values) between studies focussing on specific risk markers, including gender (self-injury & aggression), IQ (self-injury, aggression & destruction), AS (aggression), Autism (self-injury), CdLS (self-injury), FXS (self-injury & aggression), PWS (aggression) and SMS (self-injury). This suggests considerable difference in the way the included studies review either the behaviour that challenges or the risk marker, or differences in the sample characteristics, such as age, proportion of a particular gender or severity of ID. The resultant impact is that the RR findings need to be interpreted with significant caution.

Age represents an interesting methodological variation characteristic to consider for heterogeneity. Shanahan et al. (2008) indicated that behavioural profiles differ significantly between younger and older individuals with FXS. Interestingly, one of the studies excluded in the self-injury-FXS analysis had a higher mean sample age than the other studies included. Alternatively, this meta-analysis has indicated that individuals with severe/profound ID are more likely to engage in any one of the three forms of behaviour that challenges; comparison

of two studies with samples differing in severity of ID could create heterogeneity. This could also occur when comparing predominantly male and female samples.

Consideration should also be made for lack of unanimity in control populations; there is a trend to select Down syndrome as a control population, however Down syndrome has been determined to be a 'preventative' risk marker, thus comparison with this population will result in over-estimation of syndrome RR significance. Moreover, heterogeneity could occur when studies compare a syndrome to a different control population, e.g. one comparing to Down Syndrome and another comparing to mixed aetiology ID.

Appendix A indicates the array of measures used to determine presence of self-injury, aggression and destruction, which include the Modified Overt Aggression Scale (Oliver et al., 2007), Challenging Behaviour Questionnaire (Hyman et al., 2002), Behaviour Problem Inventory (Rojahn et al., 2001), Self-injury, Aggression and Destruction Screening Questionnaire (Davies & Oliver, 2016) and a number of self-determined measures. The discrepancies among these measures could result in considerable heterogeneity in results. The measures vary between the behaviour having to have occurred within the last week (Modified Overt Aggression Scale), to the last 6 months (Developmental Behaviour Checklist) or even occurred ever, with frequency being used to determine whether the behaviour is present (SAD-SQ; Bhaumik et al. (1997) self-developed measure). Measures focussing on the last week are more likely to identify individuals with forms of behaviour that occur more frequently, whilst measures focussing on the last month could be identifying much more rare/infrequent behaviours.

Generally, measures of self-injury are consistent in their definition that it is a self-directed, harmful behaviour. Discrepancies arise, however, for definitions of aggression and destruction. The BPI adopts a subscale of aggressive/destructive behaviour, with property destruction,

pushing and verbal outbursts all being part of the same subscale. In contrast, the MOAS fragments the term 'aggression' into verbal aggression, physical aggression, property aggression and self-aggression. The CBQ and SAD-SQ also consider subtypes of behaviour in this way. This approach provides a much richer insight into the specific topographies of different socially maladaptive behaviours. A review of aggressive behaviour in ID research by Benson & Brooks (2008) indicated a need for assessment instruments that address severity and topography of aggression in isolation. As previously indicated, data relating to aggression obtained using the BPI was excluded from this meta-analysis as it was not possible to remove incidences of destruction from the subscale. Based on the current meta-analysis, it is clear that there is insufficient knowledge regarding differences between aggressive and destructive behaviour and that condensing the two into one subscale is limiting the potential pool of knowledge regarding aggression and destruction.

1.6.6 Conclusion

In summary, this meta-analysis sought to identify demographic risk markers, specific syndromes or diagnoses and other risk markers that impact the risk of self-injury, aggression and destruction. Whilst it has made a comprehensive advance in achieving this aim, and has synthesised the available literature, there remains further advancements to be made. For example, syndromes such as Cri du Chat, Sotos and Rett syndrome have been associated with increased self-injury (Collins and Cornish, 2002; Sheth et al., 2015; Oliver et al., 1993), however insufficient data were available to conduct a meta-analysis to determine the impact of such syndromes. As the available data increases, it would be valuable to continue to quantitatively calculate the risk these syndromes present, as well as consistently re-evaluating the analyses presented based on a small number of syndromes. Furthermore, improved focus on aggression and destruction as two separate behaviours, adopting measures that embrace this concept, will facilitate more advanced understanding of aggression and destruction and

determination of whether they have different risk markers. Clinically, the results of this meta-analysis could be translated to aid identification of individuals that are at elevated risk of developing a specific form of behaviour that challenges, based on the risk markers they present, and could facilitate development of early-identification and intervention strategies.

CHAPTER 2

Clinical utility of risk markers for aggression and destruction in children with intellectual disability

2.1 Abstract

Rationale: The adverse effects of behaviours that challenge are vast, including reduced quality of life, institutionalisation, negative carer attitudes and financial cost. This study sought to determine if it is viable to consider an early intervention route, similar to that used in psychosis, for aggression and destruction in children with intellectual disability. **Method:** Using a large data pool as a modelling sample, a backward stepwise logistic regression was used to develop an algorithm that combined risk markers that most accurately predicted current presence or absence of aggression and destruction. An optimal cut off to maximise sensitivity whilst maintaining good specificity was established. This algorithm was then applied to a new test set of data for cross validation. **Results:** The most predictive risk markers included in the aggression algorithm were gender, autism, skin problems, health problems, overactivity, impulsivity and repetitive behaviour. This model achieved a sensitivity of 83.2% and specificity of 70.2% in the test sample. Presence of autism and impulsivity were the most significant predictors in the destruction algorithm, giving a sensitivity of 80.5% and specificity of 63.8% in the test sample. **Conclusion:** This study demonstrates that it is possible to develop an algorithm that predicts currently present behaviour. The next stage of considering early intervention for aggression and destruction will be longitudinal prediction studies.

2.2. Introduction

2.2.1 Behaviours that challenge

'Behaviours that challenge[8]' are a clinical concern for many individuals and families, with estimates of such behaviour in typically developing adolescents ranging from 5% to 20% (Marsee et al., 2014; Raine et al., 2006). The term 'behaviours that challenge' refers to any behaviour that presents a significant challenge to services, the individual, or those around them, regardless of the presumed cause of the problem. Among children with intellectual disability (ID), such behaviours typically comprise aggression, destruction of property (destruction) and self-injury (Lowe et al., 2007). However in typically developing children, the spectrum of behaviours is far broader, incorporating drug abuse, sexual promiscuity and vandalism (Newcomb & Bentler, 1989). 'Aggression', as defined by the Royal College of Psychiatrists (2007), includes physical aggression, verbal aggression and sometimes property destruction. Physical aggression can be used to refer to an array of acts, including physical assaults on peers, family members or staff, such as pushing, kicking or biting (Rojahn et al., 2001). 'Destruction' refers specifically to the intentional damage of property, such as breaking an object into two or more pieces, ripping things from walls or into parts, or denting/marking solid objects (Emerson et al., 2001). Of the proportion of typically developing children that engage in some form of aggressive behaviour (including destruction), only 5-10% are likely to continue this behaviour into adulthood (Moffitt, 1993; Tremblay et al., 2004) and only 5% of those with extreme behaviour in childhood are likely to experience adverse impacts, such as drug addiction and criminality by age 25 (Fergusson et al., 2005). In contrast, individuals with ID that engage in behaviours that challenge are more likely to experience reactive physical intervention, psychiatric hospital intervention, school exclusion and decreased quality of life (Beadle-

[8] The term 'behaviours that challenge' has been used instead of 'challenging behaviour' in line with adjustments in terminology within this field to reduce implications of blame towards the individual engaging in such behaviour.

Brown, Murphy & DiTerlizzi, 2009; Mandell, 2008; Allen, Lower, Brophy & Moore, 2009; Knapp et al., 2005). Moreover, staff members working with individuals showing behaviours that challenge are more likely to hold negative views or be more critical towards those showing such behaviour (Weigel, Langdon, Collins & O'Brien, 2006). This suggests there is considerable benefit in focusing research efforts on better understanding behaviours that challenge in the ID population, as they experience more adverse effects as a result.

The prevalence of behaviours that challenge observed among individuals with intellectual disabilities (ID), autism and learning difficulties is significantly greater than that observed in typically developing children (21% compared to 4% in Britain specifically; Emerson & Hatton, 2007). When individual topographies of behaviours that challenge are considered, specifically aggression and destruction, the prevalence is highly variable, ranging from 10% to 77% (Crocker et al., 2006; Emerson et al., 2001; Tyrer et al., 2006; Emerson et al., 2001). Aggressive behaviour is most common, with most estimates clustering around 30-40% (Davies & Oliver, 2016; Lowe et al., 2007; Powis & Oliver, 2014) whilst the estimated prevalence of destructive behaviour is between 15-40% (Ruddick et al., 2015; Davies & Oliver, 2016). Aggressive and destructive behaviours are reported to emerge early in life for people with ID, with prevalence of behaviour problems ranging from 20-64% in preschool age children (Roberts, Mazzucchelli, Taylor & Reid, 2010). Furthermore, in contrast to the small proportion of typically developing children that are likely to continue to engage in behaviours that challenge into adulthood, behaviours that challenge are shown to be highly persistent among people with ID (Totsika & Hastings, 2009) and consistent in epidemiology (Davies & Oliver, 2016). The impact of such persistent, early emerging behaviours that challenge is substantive. In addition to the consequences to the individual, behaviours that challenge result in high financial cost to services; cost to services for adults with severe behaviours that challenge are estimated between £89,335 and £358,415 (Cooper, Emerson, Glover & Gore, 2014). Due to the adverse impacts

of these behaviours, it is necessary to devise comprehensive treatment approaches to ameliorate them, as once behaviours enter the repertoire, without treatment, problematic behaviour is likely to escalate (Emerson et al., 2001).

2.2.2 Treatment approaches

Operantly informed treatments emerged following empirical demonstration that presence and severity of these behaviours is influenced by social negative reinforcement, social positive reinforcement and sensory reinforcement (Carr, 1977). Studies have demonstrated that contingent delivery of a stimulus that is socially mediated (attention, preferred activities or tangibles) contributes to maintenance of aggression or destructive behaviour (Richman & Hagopian, 1999; Ringdahl et al., 2018). Similarly, social negative reinforcement may maintain behaviours that challenge though removal of socially negative reinforcers, such as task demands, on a contingent basis (Carr & Durand, 1985). A number of interventions based on these hypotheses have been devised, including differential reinforcement (reinforcement contingent on presence of alternative behaviour), noncontingent reinforcement (reinforcement based on a time schedule to reduce behaviours dependent on non-social reinforcement or socially determined consequences) and antecedent interventions (reducing aversiveness of a task by developing more adaptive skills). These strategies are effective among both children and adults with ID (Catania, 2013; Chowdury & Benson, 2011; Lindberg et al., 2003; Laraway et al., 2003; Shogren et al., 2004; Romaniuk & Miltenberger, 2001). Positive Behaviour Support (PBS) offers a combination of the conceptual framework of applied behaviour analysis within a person-centred approach. PBS has been indicated as effective in minimising behaviours that challenge (MacDonald & McGill, 2013; Langdon et al., 2017). Whilst these interventions are effective, they are responsive, and often expensive.

One important limitation of current applications of differential and noncontingent reinforcement, and antecedent interventions is that they typically depend on the behaviour

already being present and the individual coming into contact with appropriate specialised, high-cost services (Feldman et al., 2004). Moreover, there is risk of the behaviour re-emerging if the reinforcement schedule is not maintained; successful interventions often necessitate highly controlled environments and precise management of environmental contingencies (Goh et al., 2000; Berg et al., 2007; Wacker et al., 2011). Thus, an alternative, complementary approach for these behaviours is to adopt an early intervention pathway (Emerson, 1995), similar to that in psychosis and dementia.

2.2.3 Early-intervention

Early intervention is characterised as an attempt to delay or prevent psychopathology by offering an intervention prior to symptoms becoming clinically significant (Ramey & Ramey, 1998). Early intervention has proven effective for treatment of psychosis; randomised control trials have shown positive results from identifying and treating people in the prodrome phase (those with no symptoms of psychosis but considered at high risk of developing psychosis), with reduced chances of those treated transitioning to psychosis (McGorry et al., 2002). Moreover, early psychosis services are often more cost effective than standard services as there is less need for costly inpatient care (McCrone et al., 2009).

The need for early recognition of behaviours that challenge has long been recognised due to their association with childhood onset and significant adverse effects (Emerson, 1995). Despite research indicating that risk of developing behaviours that challenge emerges early in childhood (Totsika et al., 2011; Einfeld et al., 2010), only a small volume of literature has sought to examine the practicality of early intervention (Cooper et al., 2014). This research has shown positive outcomes through skill replacement, derived from functional analysis, when implemented early (Harvey et al., 2009). There is a clear rationale for interventions to reduce the likelihood of children developing a repertoire of aggressive or destructive behaviours. It is not financially viable, however, in the current healthcare climate to offer *all* children such

intervention approaches. Systematic early identification of at-risk individuals and rapid response provides an alternative, more affordable route to ensuring behaviours do not develop. Chapter 1 demonstrates that there is a notable lack of research regarding risk markers (variables that correlate) for aggression, and an even greater lack of research on risk markers for destruction. Identification of risk markers for these specific behaviours that challenge would highlight targets for intervention strategies (for example, if communication deficits represented a risk marker for behaviours that challenge, interventions could focus on improving communication skills). Improved knowledge on risk markers for these behaviours would confer the opportunity to identify children before aggression/destruction occurs, to enable early intervention.

2.2.4 Risk markers of aggression and destruction

Literature concerning risk markers for self-injury is relatively robust, however the meta-analysis outlined in Chapter 1 indicated that there is significantly less research into risk markers for aggression and destruction. Chapter 1 identified a number of significant risk markers for aggression (severity of intellectual disability, living in paid or congregate care, presence of autism, presence of Smith Magenis Syndrome and deficits in expressive communication, with Down Syndrome representing a protective marker). Additional genetic syndromes have been noted as risk markers by individual studies, such as Angelman Syndrome (Arron et al., 2011), as well as overactive or impulsive behaviour (Davies & Oliver, 2016), however an insufficient number of studies have presented findings to evaluate this reliably and this represents an important area for future exploration. The literature regarding destruction, however, was relatively limited. The results in Chapter 1 highlighted methodological concerns, where many studies conflated measurement of aggression and destruction, making it different to isolate factors that contribute solely to destruction. Thus, only intellectual disability and presence of autism were identified as risk markers for destructive behaviour; however, individual empirical

studies have highlighted age (Chadwick et al., 2000; Lowe et al., 2007), mobility (Chadwick et al., 2000), repetitive behaviours and impulsivity (Davies & Oliver, 2016) as markers of significant interest. Chapter 1 highlighted the need for further research in the areas of aggressive and destructive behaviour to contribute to the limited knowledge base.

Giving professionals (Health Visitors, GPs, Early Years Practitioners) the tools to identify individuals with risk markers associated with increased risk of aggressive or destructive behaviour could facilitate earlier intervention, with the hope of reducing the likelihood of these behaviours developing and could help to determine children that would most benefit from a proposed intervention (Cooper et al., 2014). There has currently, however, been 1) poor identification of risk markers for aggression and destruction of property specifically, and 2) no translation of the known risk markers into a clinically viable tool that can predict which children would benefit most from early intervention. The Self-injury, Aggression and Destruction Screening Questionnaire (Revised; SAD-SQ(R)) was devised for the purpose of this research based on the SAD-SQ (Davies & Oliver, 2016). It incorporates risk markers thought to be associated with behaviours that challenge and is sufficiently brief for clinical application. The present study seeks to determine whether components of the SAD-SQ(R) could act as a low-cost and effective screening tool for identifying the risk of developing aggressive or destructive behaviour.

2.2.5 Aims of current study

The current study has the following aims:

1) To identify and quantify the contribution of risk markers to the presence of aggressive and destructive behaviour
2) To develop an algorithm capable of predicting current presence or absence of aggression and destruction.

3) To test the developed algorithm in a new data set to determine if this strategy has potential clinical utility to identify individuals who have risk markers that are suggestive of increased risk of aggressive or destructive behaviour.

2.3. Method

2.3.1 Ethics

Ethical approval to complete the current study was provided by the North East Tyne & Wear South Research Ethics Committee. The Health Research Authority Number was: 235418 (Protocol Number: RG_17 182; REC Number 18/NE/0249; Appendix C). Participants were contacted (Appendix D) and given sufficient time to review the information (Appendix E) prior to giving consent (Appendix F) and were welcome to take the information home to discuss with family. For details on data protection, potential distress and support offered, please review the participant information sheet (Appendix G).

2.3.2 Recruitment

2.3.2.1 Modelling Sample

The first stage of the research utilised a rich set of pooled data from three previously published studies (Richards, Davies & Oliver, 2017; Davies & Oliver, 2016; Handley, 2014). The methodology for the collection of these data is reported in more detail in these publications. Handley (2014) recruited children with severe or profound intellectual disabilities aged 2-11 attending a Child Development Centre, assessment group or school for children with intellectual disabilities in Birmingham. Richards et al. (2017) recruited 515 children and adults with a diagnosis of autism from schools and adult services linked to the National Autistic Society, whilst Davies and Oliver (2016) recruited 629 children diagnosed with a severe intellectual disability aged 2-12 from special schools in the Birmingham area. The total modelling sample therefore included 1550 participants, before cleaning.

2.3.2.2 Test Sample

Recruitment of the test sample utilised three pathways to recruit a representative sample of children with neurodevelopmental disability or developmental delay from 1) a previously established participant database; 2) special education schools in the West midlands; 3) health services in the west midlands (including school nurses, child development centres, community paediatricians and health visitors. Table 2.1 presents the return rates for each route.

2.3.3 Measures

The current study used a revised version of the SAD-SQ (Davies & Oliver, 2013). The SAD-SQ for the purpose of this study was revised to include questions regarding communication skills to inform future interventions, however the data was not analysed in this study. There are two versions of the SAD-SQ(R); a 54-item questionnaire for children under the age of 6 (Appendix G), and a 64-item questionnaire for those 6 and over (Appendix H). The under 6 questionnaire obtained developmental delay information in line with the Denver Developmental Screening Test II (Frankenburg, Dodds & Archer, 1990), whilst the 6 and over questionnaire used the Wessex Behaviour Scale (Kushlick et al., 1973). Two different questionnaires for different age groups were necessary as the Wessex Behaviour Scale asks about abilities that younger children would not be able to do, for example being able to read. The SAD-SQ(R) focusses on simplicity and accessibility, achieving maximum information about risk markers in minimum time. This was achieved by reviewing all questionnaires devised that gather information on the target risk markers (Davies & Oliver, 2013). The SAD-SQ(R) presents examples to aid completion and assesses health problems, ability, repetitive behaviour, level of ability and different types of behaviours that challenge (self-injury, aggression and destruction). Response scales are Likert, binary or short answer and the measure is informant report, completed by a carer or parent familiar with the child. For each behaviour that challenges, frequency (0 = never, 4 = very often), management difficulty (0 = not difficult,

4 = extremely difficult) and concern (0 = not at all concerned, 4 = extremely concerned) were assessed. The SAD-SQ is reported to have good concurrent and convergent validity, as well as good reliability (Davies & Oliver, in review). Inter-rater reliability was indicated as between .21 and.47, with overactivity/impulsivity representing the lowest reliability score, although other papers have found similar results with different measures (Amador-Campos et al., 2006). Moreover, those categorised as 'high risk' by the SAD-SQ also scored significantly higher than those categorised as 'low risk' on other standardized measures, e.g., overactivity ($U = 33$, $p = .001$), impulsiveness ($Z = - 2.727$, $p < .008$), repetitive ($U = .49$, $p = .003$) and restricted ($U = 61.5$, $p = .017$).

2.3.3.1 Classification of aggression and destruction

The SAD-SQ(R) rated severity, concern and management of aggression and destruction on scales of 0 to 4, with 0 indicating never and 4 indicating very frequently. The current study sought to identify behaviours that would be considered clinically significant; sufficiently problematic that parents would be likely to seek support. Previous studies have taken this approach, classifying 'clinically significant' behaviours as those scoring either a 3 or 4 for severity (Richards et al., 2017; Davies & Oliver, in review). In order to produce binary responses of yes/no for presence of aggression and presence of destruction, responses of 3 and 4 for severity were accepted as being indicative of behaviour likely to be deemed challenging in a clinical nature. Scores of 1 or 2 for frequency could be deemed very low-level difficult behaviour that would not reach clinical significance. In addition, however, in order to identify all clinically significant behaviour, the management and concern scores were reviewed when frequency was rated as 1 or 2. It may well be that a behaviour is infrequent, but is hard to manage when it does present, therefore this could still present a clinical concern. If both management *and* concern returned scores of 1 or 2, this behaviour was not considered 'challenging' and therefore the data were coded as non-aggressive/non-destructive. Where

either management *or* concern gave a score of 3 or 4, the behaviour was considered to be clinically significant and coded as present.

2.3.3.2 Categorising independent variables

Where possible, to support analysis of risk, predictor variables were converted into binary data.

Ability: For children under the age of six, ability was determined by summing Denver Score items (max score 20). Scores from 0-12 were considered to show a deficit in ability, whilst scores of 13 and above were considered to show no deficit. For children aged six and above, three items were added together from the Wessex Questionnaire (Can the child wash, dress and feed themselves; max score 9). Participants scoring 0-8 were considered to have a deficit in ability, whilst those scoring 9 were considered typical.

Speech: For children under age 6, the Denver item 'Can your child say two words?" was used. Only three children were one year old at the time of completion, whilst the remaining children were two years old or older. Language development literature suggests that most typically developing children are able to say two words together by 18 months and over (Luinge et al., 2006), therefore this was considered an appropriate speech categorisation. If they could say two words, they were identified as having typical speech, whilst they were identified as having a speech deficit if they could not. For children age six and above, the Wessex question relating to speech was used, with responses of 'never says a word' and 'odd words only' being indicative of a speech deficit.

Ordinal factors: Frequency of repetitive behaviour and frequency of obsessive/ritualistic behaviour remained ordinal scores (rated 0, never, to 4, extremely frequently). Impulsivity was calculated by adding together 'does the individual find it difficult to wait?' (rated 0, never, to 4, extremely frequently) and 'does the individual want things immediately?'(rated 0, never, to 4, extremely frequently). Overactivity was calculated by adding 'does the individual act as if

driven by a motor?' (rated 0, never, to 4, extremely frequently) and 'does the individual want things immediately?' (rated 0, never, to 4, extremely frequently). Impulsivity and overactivity scores remained as scale scores as there is no clear cut off for what constitutes a clinical score for overactivity or impulsivity. This was scored in line with the validity paper on the SAD-SQ (Davies & Oliver, 2016).

Health factors: For vision and hearing, if participants were blind/had poor sight or deaf/had poor sight, they were considered to have a problem in this area. For eye, skin, respiratory, dental, digestive and ear problems, a score of never or mild was considered to be indicative of no problem, whilst a score of severe or moderate was indicative of a problem being present.

2.3.4 Procedure

The current study was part of a wider, longitudinal research study. Participants included in the current study were recruited for the first stage (Time 1) of the study. Participation in the study equated to a maximum of 45 minutes of participant time; 30 minutes to carefully read the information pack and provide informed consent, 15 minutes to complete the questionnaire.

Participants held on the Cerebra Centre database previously consented to be contacted regarding future research. To avoid priming, the study was described as aiming to 'better understand the various behaviours of children with neurodevelopmental disabilities (Appendix E). An information pack was sent directly to eligible individuals with an opt-in consent form. 181 participants were contacted through the database recruitment stream. If the team had not received a questionnaire response within two weeks, a follow up letter was sent. The option to compete the questionnaire over the phone was given, or for a researcher to visit their home to complete the questionnaire (within 10 miles of the university base). This stream returned 45 questionnaires.

Special schools in the West Midlands area were contacted by phone or email and provided with information packs about the study. Consent was required from the Head Teachers or leads of schools and nurseries for their students to participate. 928 information packs for parents/carers were distributed via schools and placed in the book-bags of all pupils with developmental delay that met the age inclusion criteria. Reminder flyers were also provided to schools to be placed in book-bags two weeks after distribution of the information pack. 86 participants were recruited through this recruitment stream.

For the third recruitment stream via the NHS, an invitation letter was distributed with routine pre-clinic appointment letters or by research nurses indicating that the research team would be present in the clinic. Research teams attended Child Development Clinics (CDCs) in the West Midlands and healthcare professionals were proactive in identifying individuals that met inclusion criteria directing them to the research team. These individuals were offered the same information pack previously described. Written consent was obtained from carers of eligible participants in the waiting room or in private rooms where available. If parents or carers expressed an interest but were not able to complete the questionnaire in the clinic, they were given a pack containing a pre-paid envelope to take home. Contact details were taken if the potential participant was happy to disclose, and they were contacted 2 weeks later for follow-up if no questionnaire form was received within that time. Again, questionnaires could be completed over the phone or visits within 10 miles arranged. 1765 packs were distributed through this stream and 194 were completed.

Table 2.1 Return rates for each recruitment stream

	Packs sent	Packs completed	Response rate
Clinical	1765	194	11.0%
School	928	86	9.3%
Existing database	181	45	24.9%

Due to multiple recruitment streams being used, there was a risk of participants being contacted multiple times, therefore all invitation letters indicated this risk and asked carers to disregard the information if they had already been contacted. All individuals in contact with researchers in this study, regardless of whether they chose to participate, were offered signposting to the FIND Website (www.findresources.co.uk), which contains helpful information about the difficulties children and their parents may face following diagnosis of developmental delay.

2.3.5 Inclusion criteria

Participants were eligible to take part in the study if they were aged between 2 and 11 years, thought to have a developmental delay, autism or genetic syndrome. Attending paediatric clinics for children with suspected neurodevelopmental disability or developmental delay, or a school for children with developmental delay was considered sufficient evidence for the child to meet this criterion. In addition, parents or carers needed to be both cognitively and physically able to complete the assessment and have necessary language skills to give informed consent and understand the questionnaire. Finally, the child and carer must live in the West Midlands or be part of a research participant database for children with rare genetic syndromes.

Burderer's (1996) methodology was utilised to calculate the target sample size for adequate (90%) specificity and sensitivity, which resulted in 346 participants being required. This was, however, on the basis of the larger study, which was longitudinal and had to account for attrition between Time 1 and Time 2. 325 participants aged between 2 and 11 years of age were

recruited via the three recruitment pathways who had either confirmed or suspected developmental delay. The slightly lower recruitment compared to target was due to delays in obtaining ethical approval, and for the longitudinal study, could slightly increase the risk of Type II error, however should not have any adverse impact on power for analyses in the present study (Moher et al., 1994).

2.3.6 Participants and analysis approach

The current study necessitated the use of an initial sample (preliminary sample) obtained from the compilation of data from the three previous studies. This sample includes data on every covariate included in the preliminary sample data. Due to the number of participants needed for the algorithm analysis to be effective (as indicated by a power analysis), covariates which had low response rates had to be removed, thus, a subset of the modelling sample was used for the development of the algorithm (Subset of Modelling Sample). During the regression analysis later conducted, covariates of interest were identified. In order to maximise the number of participants included in the development of the algorithm, a second modelling sample was created (Modelling Sample), removing covariates that were not of interest. Due to poor response rates on some of the removed covariates, this greatly increased the number of participants that informed the algorithm, improving the robustness of the subsequent data. To aid transparency in the data included in the study, the following sections will detail each of these samples and the processes involved in their compilation.

2.3.6.1 Developing Subset of Modelling Sample

Subset of Modelling Sample is based on the complete set of data obtained from the compilation of data from the three outlined studies.

Aggression Subset of Modelling Sample: 1531 participants provided a response to aggression frequency that enabled classification of presence or absence of aggression. This is known as

the preliminary Aggression Modelling Sample. Based on the methodology presented by Peduzzi et al. (1996), the minimum number of cases necessary for logistic regression was calculated using the following equation:

$$N = (10 * K) / P$$

Where *K* = the number of covariates being assessed and *P* = the proportion of positive cases of the behaviour in the population. To provide an example; if a sample had 100 participants and 20 showed self-injury, the proportion of positive cases would be 20%. If there were four covariates, K would be 4. N = (10*4) / 0.20; 200 participants would be needed.

For the aggression sample, there were 20 covariates and the proportion of aggression was 27%: *N = (10*20)/0.27 = 740*. When all variables were included in the analysis, a backward conditional logistic regression using the preliminary Aggression Modelling Sample only resulted in 146 cases being included in the analysis, due to missing data. This was an insufficient number of cases. Conditional regression has multiple options for missing data; exclude the participant, replace the missing data with a mean based on the present data or weight the cases to bias those with data entered (Raghunathan, 2004). As the intention of this analysis is to develop an algorithm, exclusion was the most viable option to maintain robustness. Missing data were reviewed for each variable. Table 2.2 provides an overview of complete data for each variable obtained from the SAD-SQ(R). In the preliminary Aggression Modelling Sample, two variables had over 60% data missing (epilepsy and respiratory problems) and an additional variable had over 45% data missing (genetic syndrome).

Table 2.2 Summary of % complete responses for each variable in SAD-SQ(R) for Aggression and Destruction preliminary Modelling Samples

Variable	Preliminary Aggression Modelling Sample (N=1531)		Preliminary Destruction Modelling Sample (N = 1540)	
	N Valid	% of N	N Valid	% of N
Age	1464	95.6	1471	95.5
Gender	1497	97.8	1504	97.7
Autism	1333	87.1	1341	87.1
Genetic Syndrome	994	64.9	1004	65.2
Vision problems	1088	71.1	1094	71.0
Hearing problems	1078	70.4	1083	70.3
Eye problems	1467	95.8	1472	95.6
Ear problems	1477	96.5	1483	96.3
Dental problems	1480	96.7	1486	96.5
Digestive problems	1475	96.3	1481	96.2
Skin problems	1491	97.4	1497	97.2
Respiratory problems	464	30.3	466	30.3
Epilepsy	462	30.2	464	30.1
Number of health problems	1500	98.0	1506	97.8
Freq of Repetitive behaviour	1471	96.1	1474	95.7
Obsessions/rituals	1472	96.1	1477	95.9
Impulsivity	1477	96.5	1483	96.3
Overactivity	1431	93.5	1437	93.3
Speech deficit	1056	69.0	1061	68.9
Ability deficit	1036	67.7	1040	67.5

It was decided to exclude any factors that had less than 65% of completed cases. This resulted in the exclusion of epilepsy, respiratory problems and genetic syndromes and changed the minimum participant formula to: $N = (10*17)/0.30^{9} = 531$. Following the exclusion of the above 3 variables, 631 (41.2%) participants had complete data across all factors to be included in the regression. The demographics of this sample, Subset of Modelling Sample, are shown in Table 2.3.

Destruction Subset of Modelling Sample: 1540 participants provided a response to destruction frequency that enabled the classification of presence or absence of destruction. Based on the calculation by Peduzzi et al. (1996), the minimum number of cases for the destruction model

[9] Formula adjusted in line with new prevalence using 17 variables

regression was: $N = (10*20)/0.21 = 952$. Only 147 participants had sufficient data for regression analysis in the preliminary Destruction Modelling Sample. As with the preliminary Aggression Modelling Sample, both respiratory problems and epilepsy had less than 35% complete responses (see Table 2.2). Exclusion of respiratory problems, epilepsy and genetic syndromes, due to having less than 65% complete data, resulted in 633 participants having sufficient data for regression. This did not meet the minimum threshold criteria for 17 variables: $N = (10*17)/0.23^{10} = 739$, however the remaining variables with a higher proportion of missing data (ability and speech) were considered important to include, based on the literature review, as these have been identified as potential risk markers, at the potential expense of power/ increase of Type 2 error. This is referred to as Destruction Subset of Modelling Sample (Table 2.3).

Table 2.3 Demographic information for aggression and destruction Modelling Samples 1

	Aggression Subset of Modelling Sample	Destruction Modelling Sample 1
Total Sample	N=631	N=633
Mean Age (SD)	15.8 (12.4)	15.7 (12.4)
N Male (%)	460 (72.9%)	462 (73.0%)
Autism Diagnosis (%)	481 (76.2%)	481 (76.0%)
Visual problems	117 (18.5%)	117 (18.5%)
Hearing Problems	36 (5.7%)	36 (5.7%)
Eye Problems	0 (0.0%)	0 (0.0%)
Ear Problems	16 (2.5%)	16 (2.5%)
Dental Problems	27 (4.3%)	27 (4.3%)
Digestive Problems	35 (5.5%)	35 (5.5%)
Skin Problems	42 (6.7%)	41 (6.5%)
Med. number of health problems (IQR)	1 (1)	0 (1)
Med. Frequency repetitive behaviour (IQR)	1 (2)	1 (2)
Med. Frequency obsessive behaviour (IQR)	2 (3)	2 (3)
Med. Impulsivity (IQR)	2 (5)	2 (5)
Med. Overactivity (IQR)	1 (3)	1 (3)
Speech deficit	376 (59.6%)	379 (59.9%)
Ability deficit	348 (55.2%)	349 (55.1)

2.3.6.2 Modelling Sample

After the identification of the risk markers to be included in the aggression algorithm (Section 2.4.2), participants that were previously excluded from analysis due to missing data in one of the variables not selected in the model no longer had incomplete data. This resulted in an

[10] [10] Formula adjusted in line with new prevalence using 17 variables

increase in participants available for optimising the model (Section 2.4.3). The demographic information of the participants (N=1124) included in this stage of analysis, Aggression Modelling Sample, are described in Table 2.4. This process was replicated for the destruction data set, resulting in an increase to 1307 participants included in Destruction Modelling Sample (see Table 2.4 for a description of the participant characteristics).

2.3.6.3 Test Sample

Aggression: The test sample initially comprised 325 participants. Following the exclusion of participants with missing data for the variables in the relevant devised algorithm, 284 participants remained in the Aggression Test Sample and 310 remained in the Destruction Test Sample. Table 2.4 presents the characteristics of these samples.

Tests for statistical difference between the Modelling and Test samples were conducted (Table 2.4). For non-parametric variables, median and interquartile range (IQR) were reported and independent samples Mann-Whitney U tests (z) were conducted. For categorical variables, Chi Square (X^2) tests were conducted.

Table 2.4 indicates that the majority of variables are significantly different between the two samples for aggression, with only gender (X^2=1.262, p=.261) and presence of autism (X^2=.004, p=.950) showing no significant difference across samples. The Aggression Test Sample has a significantly lower mean age (z= -6.202, p=<.001). Table 2.4 indicates that gender (X^2=.020, p=.888) and diagnosis of autism (X^2=.154, p=.695) were the only variables to not be significantly different between the two samples for destruction. The remaining variables all showed significant difference at the p<.001 level. Due to the intention of this study being test whether an algorithm can be created and be effective across different samples, this was not considered a problem; the Test Sample is representative of the sample a prospective

intervention/ need to identify individuals would be used in, whilst the Modelling Sample provides a more diverse range of individuals upon which to shape the model.

Table 2.4 Statistical difference between Aggression Modelling Sample and Aggression Test Sample and statistical difference between Destruction Modelling Sample and Destruction Test Sample

	Aggression Modelling Sample N=1124	Aggression Test Sample N=284	*z*	X^2	*p*	Destruction Modelling Sample N=1307	Destruction Test Sample N=310	*z*	x^2	*p*
N aggression (%)	339 (30.2%)	154 (54.2%)	-	58.428	<.001*	302 (23.1%)	146 (47.1%)	-	72.020	<.001*
Mean age (SD)	11.9 (11.4)	6.7 (2.6)	-6.202	-	<.001*	11.9 (11.4)	6.7 (2.6)	-6.430	-	<.001*
Males (%)	827 (74.0%)	201 (70.7%)	-	1.262	.261	932 (71.3%)	220 (70.9%)	-	.020	.888
Autism diagnosis (%)	736 (65.0%)	184 (64.8%)	-	.004	.950	853 (62.3%)	197 (63.5%)	-	.154	.695
Med. Impulsivity score (IQR)	4 (5)	6 (4)	-10.024	-	<.001*	3 (5)	6 (4)	-10.790	-	<.001*
Med. Overactivity score (IQR)	2 (5)	5 (5)	-8.400	-	<.001*	2 (5)	5 (5)	-9.268	-	<.001*
Med. Repetitive behaviour score (IQR)	1 (3)	3 (3)	-5.230	-	<.001*	1 (3)	3 (3)	-4.909	-	<.001*
Med. Obsessive behaviour score (IQR)	2 (3)	3 (3)	-3.507	-	<.001*	2 (3)	3 (3)	-3.188	-	.001*
Med. number health problems (IQR)	1 (2)	1 (2)	-9.691	-	<.001*	1 (2)	2 (2)	-9.316	-	<.001*
Ability deficit (%)	489 (43.5%)	157 (55.3%)	-	12.704	<.001*	469 (35.9%)	174 (56.1%)	-	42.656	<.001*
Speech deficit (%)	506 (45.0%)	38 (13.4%)	-	95.430	<.001*	512 (39.2%)	101 (32.6%)	-	4.633	.031*
Skin Problems (%)	107 (9.5%)	59 (20.7%)	-	27.388	<.001*	137 (10.5%)	64 (20.6)	-	23.458	<.001*

2.3.7 Creating a new risk marker

To identify variables associated with aggression and destruction, the variables obtained from the SAD-SQ(R) (Subset of Modelling Sample) were entered into a backward stepwise logistic regression. Aggression and destruction were examined separately, and therefore each was the dependent variable in the respective analysis. Following this, a new variable was calculated using the algorithm developed from Subset of Modelling Sample. This variable was then manipulated into an S curve and an optimal cut off determined using Modelling Sample. The same variable was then calculated in the Test Sample, and cut offs explored to determine if they were replicable of the cut-off identified in Modelling Sample.

2.4 Results

2.4.1 Appraising current risk markers

To identify and quantify the contribution of risk markers to the presence of aggressive and destructive behaviour (Aim 1), analyses were conducted comparing risk markers between children with and without aggression and destruction in the modelling sample. Tables 2.5 & 2.6 present these data. For categorical risk markers (gender, speech deficit, autism, skin problems, genetic syndromes, vision problems, hearing problems, ear problems, dental problems, digestive problems and epilepsy), independent samples Chi Square tests were performed to determine whether there was a statistically significant difference between groups in terms of the presence of aggression and destruction respectively. For scale risk markers (age, repetitive behaviour, obsessive behaviour, impulsivity, overactivity and number of health problems), tests for normality indicated all data were not normally distributed, and therefore Mann-Whitney U tests were conducted to compare median scores for behaviour present and absent groups. Relative Risk (RR) for continuous data was calculated by standardizing *z* to Cohens *d*, then converting Cohens *d* to RR using the methodology outlined in Bornstein (2009). Standard Error was calculated for the variable to enable calculation of confidence intervals (CI) for the RR and determine significance.

Increased frequency of repetitive behaviour, higher impulsivity, higher overactivity, higher obsessions and presence of autism were identified as increasing risk of both aggression and destruction. Comparisons in the Aggression Modelling Sample also highlighted increased risk associated with health problems, male gender and epilepsy. Comparisons in the Destruction Modelling Sample identified speech problems, health problems, and ear problems as significant contributors to increased risk of destruction. Bonferroni correction was used when using multiple comparisons (Shaffer, 1995).

Table 2.5 Overview of risk markers and associated difference in present and absent aggression groups (Aggression Modelling Sample)

Variable		Present (N=339)	Absent (N=785)	Independent Samples Tests X^2	z	p	Effect Size RR (95% CI)	p
Age	*Mean (SD)*	10.22 (10.3)	12.6 (11.7)	-	-3.782	<.001*	1.46 (0.745, 2.85)	.263
Repetitive	*Med. (IQR)*	2 (3)	1 (3)	-	-5.601	<.001*	1.77(1.60, 1.95)	<.001*
Impulsivity	*Med. (IQR)*	7 (3)	2 (5)	-	-17.017	<.001*	6.93 (5.83, 8.30)	<.001*
Overactivity	*Med. (IQR)*	6 (5)	2 (3)		-15.121	<.001*	5.31 (4.54, 6.21)	<.001*
Obsessions	*Med. (IQR)*	3 (2)	1 (2)	-	10.634	<.001*	3.06 (2.79, 3.38)	<.001*
Health problem	*Med. (IQR)*	1 (2)	1 (1)	-	-3.909	<.001*	1.49 (1.41, 1.58)	<.001*
Speech deficit	*Yes*	98	344	1.188	-	.276	1.17 (0.87, 1.57)	.286
	No	54	233					
Gender	*Male*	282	545	23.054	-	<.001*	1.77 (1.37, 2.28)	<.001*
	Female	57	240					
Ability deficit	*Yes*	99	306	8.048	-	.005*	1.54 (1.12, 2.10)	.007*
	No	50	266					
ASD	*Yes*	256	480	21.628	-	<.001*	1.632 (1.32, 2.03)	<.001*
	No	83	305					
Skin Problems	*Yes*	44	63	6.746	-	<.001*	1.17 (0.90, 1.51)	2.18
	No	295	722					
Genetic Syndrome	*Yes*	16	72	11.303	-	.004*	0.53 (0.34, 0.84)	.006*
	No	240	460					
Vision problem	*Yes*	21	116	4.035	-	.133	0.71 (0.46, 1.10)	.122
	No	132	478					
Hearing problem	*Yes*	3	41	5.393	-	.020*	0.32 (0.11, 0.96)	.042*
	No	150	551					
Ear problems	*Yes*	29	33	8.831	-	.003*	1.62 (1.23, 2.13)	<.001*
	No	304	745					
Dental problem	*Yes*	22	40	.960	-	.327	1.20 (0.84, 1.70)	.317
	No	311	739					
Digestive problem	*Yes*	39	57	8.005	-	0.18	1.40 (1.09, 1.81)	.009*
	No	294	721					
Epilepsy	*Yes*	24	31	7.038	-	.008	1.67 (1.17, 2.37)	.004*
	No	88	248					

Table 2.6 Overview of risk markers and associated difference in present and absent destruction groups (Destruction Modelling Sample 2)

Variable	Reporting	Present (N=302)	Absent (N=1005)	Independent Samples Tests X^2	z	p	Effect Size RR	p
Age	Mean (SD)	9.8 (9.3)	12.6 (11.9)	-	-3.661	<.001*	1.45 (0.76, 2.76)	.262
Repetitive	Med. (IQR)	2 (4)	1 (3)	-	-3.982	<.001*	1.50 (1.35, 1.65)	<.001*
Impulsivity	Med. (IQR)	7 (3)	2 (5)	-	-16.382	<.001*	6.33 (5.38, 7.37)	<.001*
Overactivity	Med. (IQR)	6 (5)	2 (4)	-	-15.080	<.001*	5.29 (4.54, 6.21)	<.001*
Obsessions	Med. (IQR)	3 (2)	2 (2)	-	-8.556	<.001*	2.42 (2.19, 2.66)	<.001*
Health problems	Med. (IQR)	1 (2)	1 (2)	-	-4.134	<.001*	1.52 (1.44, 1.61)	<.001*
Speech deficit	Yes	76	437	.606	-	.463	1.49 (1.05, 2.12)	.026*
	No	42	283					
Gender	Male	239	693	9.299	-	.002*	1.46 (1.13, 1.89)	.004*
	Female	61	287					
Ability deficit	Yes	67	402	.077	-	.781	1.02 (0.73, 1.42)	.906
	No	49	311					
ASD	Yes	228	625	18.140	-	<.001*	1.63 (1.29, 2.07)	<.001*
	No	74	380					
Skin Problems	Yes	91	46	8.635	-	.003*	3.06 (2.62, 3.59)	.001*
	No	255	888					
Genetic Syndrome	Yes	21	94	6.180	-	.045*	0.70 (0.47, 1.06)	.096
	No	212	604					
Vision problems	Yes	12	153	8.326	-	.016*	0.48 (0.27, 0.84)	.011*
	No	107	595					
Hearing problems	Yes	3	51	3.591	-	.166	0.38 (0.12, 1.16)	.089
	No	118	691					
Ear problems	Yes	30	41	15.043	-	<.001*	1.90 (1.41, 2.54)	<.001*
	No	265	928					
Dental problems	Yes	23	50	2.673	-	.102	1.36 (0.96, 1.94)	.085
	No	276	917					
Respiratory problems	Yes	5	12	.798	-	.372	1.43 (0.67, 3.80)	.356
	No	84	327					
Epilepsy	Yes	7	51	3.193	-	.074	0.54 (0.26, 1.11)	.094
	No	82	285					

2.4.2 Model identification

To develop an algorithm that is able to determine current presence or absence of aggression or destruction (Aim 2), a backward stepwise logistic regression was conducted in Aggression Modelling Sample and Destruction Modelling Sample (a forward regression risks making previously included variables in the model no longer significant with each addition). Backwards was considered to be more effective. The same 17 variables were entered into the regression for each of the two behaviours: age, gender, autism, vision problems, hearing problems, eye problems, ear problems, dental problems, digestive problems, skin problems, number of health problems, frequency of repetitive behaviour, frequency of obsessive behaviour/rituals, impulsivity, overactivity, speech and ability.

Omnibus tests of model coefficients are presented in Table 2.7 (aggression) and Table 2.8 (destruction). Table 2.7 indicates that aggression was most significantly predicted by Step 9, which comprised gender, autism, skin problems, number of health problems, frequency of repetitive behaviour, frequency of obsessive behaviour/rituals, impulsivity and overactivity. Table 2.8 indicates that destruction was most significantly predicted by Step 15, which comprised autism and impulsivity.

Table 2.7: Omnibus tests of model coefficients from aggression-focussed logistic regression

Step		Chi-square	df	Sig.
Step 1a	Step	166.551	16	<.001
All variables	Model	166.551	16	<.001
Step 2b	Step	-.013	1	.911
Variable removed: Eye problems	Model	166.539	15	<.001
Step 3c	Step	-.019	1	.889
Variable removed: Age	Model	166.519	14	<.001
Step 4d	Step	-.201	1	.654
Variable removed: Digestive problems	Model	166.318	13	<.001
Step 5e	Step	-.479	1	.489
Variable removed: Dental problems	Model	165.839	12	<.001
Step 6f	Step	-.450	1	.502
Variable removed: Hearing	Model	165.389	11	<.001
Step 7g	Step	-.606	1	.436
Variable removed: Speech excluded	Model	164.783	10	<.001
Step 8h	Step	-1.909	1	.167
Variable removed: Ability + Vision	Model	162.874	9	<.001
Step 9i	Step	-2.515	1	.113
	Model	160.359	8	<.001

Table 2.8: Omnibus tests of model coefficients from destruction-focussed logistic regression

		Chi-square	df	Sig.
Step 1a	Step	90.494	16	<.001
All variables in model	Model	90.494	16	<.001
Step 2b	Step	.000	1	1.000
Variable removed: Vision	Model	90.494	15	<.001
Step 3c	Step	-.001	1	.981
Variable removed: Age	Model	90.494	14	<.001
Step 4d	Step	-.012	1	.911
Variable removed: Dental problems	Model	90.481	13	<.001
Step 5e	Step	-.036	1	.849
Variable removed: Speech	Model	90.445	12	<.001
Step 6f	Step	-.074	1	.785
Variable removed: Ear problems	Model	90.371	11	<.001
Step 7g	Step	-.339	1	.561
Variable removed: Digestive problems	Model	90.032	10	<.001
Step 8h	Step	-.426	1	.514
Variable removed: Ability	Model	89.606	9	<.001
Step 9i	Step	-.364	1	.546
Variable removed: Skin problems	Model	89.241	8	<.001
Step 10j	Step	-.377	1	.539
Variable removed: Hearing	Model	88.864	7	<.001
Step 11k	Step	-1.095	1	.295
Variable removed: Gender	Model	87.769	6	<.001
Step 12l	Step	-1.253	1	.263
Variable removed: Overactivity	Model	86.516	5	<.001
Step 13m	Step	-2.160	1	.142
Variable removed: Repetitive behaviour	Model	84.356	4	<.001
Step 14n	Step	-2.275	1	.131
Variable removed: Obsessive behaviour/rituals	Model	82.081	3	<.001
Step 15o	Step	-2.223	1	.136
	Model	79.858	2	<.001

Following the identification of the significant variables, the next stage in determining the model was to obtain the beta weights; this is the extent that each variable contributes to the model.

The significance tests and corresponding beta coefficients for the variables in the final predictive model are presented in Table 2.9 (aggression) and Table 2.10 (destruction). Table 2.9 reveals that presence of autism (*B*=1.463, *p*=.001) has the greatest weighted contribution to constant (*B*= -4.537, *p*<.001), along with impulsivity (B=.294, *p*<.001) and obsessive behaviour/rituals (*B*=.296, *p*=.026). Table 2.10 indicates that autism (*B*=1.22, *p*=.006) also had the greatest contribution of the two values included in the destruction model (*B*=-4.206, *p*<.001).

Table 2.9 Significance tests and corresponding beta coefficients for aggression risk marker model

		B	S.E	Wald	d.f	Sig.
Step 9	Gender	-.582	.302	3.700	1	.054
	Autism	1.463	.451	10.510	1	.001
	Skin Problems	-.926	.485	3.639	1	.056
	Number Health Problems	.261	.113	5.326	1	.021
	Repetitive behaviour	-.167	.090	3.479	1	.062
	Obsessions and rituals	.296	.096	9.466	1	.002
	Impulsivity	.294	.058	25.811	1	<.001
	Overactivity	.143	.064	4.975	1	.026
	Constant	-4.537	.510	79.214	1	<.001

Table 2.10: Significance tests and corresponding beta coefficients for destruction risk marker model

		B	S.E.	Wald	df	Sig.
Step 15	Autism	1.220	.448	7.422	1	.006
	Impulsivity	.359	.049	53.659	1	<.001
	Constant	-4.206	.468	80.602	1	<.001

2.4.3 Optimising the aggression and destruction models

The second stage in addressing Aim 2 was to create the predictor variable and use this to determine the optimal cut off score for classification of behaviour present. To increase statistical power, this was conducted in Modelling Sample (for further detail and discussion

please see Section 2.5.2). The new variable (*t*) was created by summing the weighted *B* values for each behaviour:

Aggression:

*t= -4.537 + (-0.582 * Gender) + (1.463 * Autism) + (-0.926 * Skin Problems) + (0.261 * Health Problems) + (-0.167 * Repetitive Behaviour) + (0.296 * Obsessions/Rituals) + (0.294 * Impulsivity) + (0.143 * Overactivity)*

Destruction:

*t= -4.206 + (1.220 * Autism) + (0.59 * Impulsivity)*

In order to achieve a prediction that is consistently contained within 1 and 0, as this enables quantification of risk in a simplified manner, it was necessary to change the exponent of the new variable from a linear projection to an S-curve. This was achieved using:

$$prediction = \frac{1}{1 + e^{-t}}$$

Where e = exponent

Logistic regressions, including that indicated above, are typically based on a cut off of 0.5 as this enables a balance between false negative/positive predictions. For the purpose of a clinical tool, however, it is more beneficial to adjust this cut off and maximise sensitivity, thereby increasing the risk of false positives. This is on the basis that individuals could miss out on beneficial interventions if the sensitivity is lowered. Thus, specificity is less essential in a clinical tool, however it is still required to prevent all individuals being categorised as 'at risk'. Therefore, a range of different cut-offs were sampled, as indicated in Tables 2.11 and 2.12. A Receiver Operating Characteristic (Area Under Curve; AUC) value was included to show the diagnostic ability of the binary classifier system.

Table 2.11 indicated that the optimal compromise of sensitivity to specificity for a screening tool for aggression, based on data in Modelling Sample, was a cut-off level of 0.27. This achieved a sensitivity of 83.2%, and a specificity of 70.2%. Overall, this model would have 74.1% overall accuracy and an AUC value of .767.

Table 2.11: Cut off exploration using Aggression Modelling Sample

Cut-off	True positive	False negative	Sensitivity	True negative	False positive	Specificity	Overall accuracy	AUC
0.1	313	26	92.3%	378	407	48.2%	61.5%	.702
0.2	291	48	85.8%	505	280	64.3%	70.8%	.751
0.25	286	53	84.4%	539	242	68.7%	73.4%	.767
0.26	283	56	83.5%	547	238	69.7%	73.8%	.766
0.27	282	57	83.2%	551	234	70.2%	74.1%	.767
0.28	279	60	82.3%	558	227	71.1%	74.5%	.767
0.29	274	65	80.8%	561	224	71.5%	74.3%	.761
0.3	273	66	80.5%	565	220	72.0%	74.6%	.763
0.4	250	89	73.7%	618	167	78.7%	77.2%	.762
0.5	216	123	63.7%	655	113	83.4%	77.5%	.736

Table 2.12 presents the different cut-offs trialled for the destruction algorithm in the Modelling Sample dataset. The results suggested that a cut-off level of 0.3 would be most beneficial. This achieved a sensitivity of 82.8% and a specificity of 69%. Overall accuracy was 72.1% and the AUC was .759.

Table 2.12: Varying cut off levels to explore sensitivity and specificity of destruction risk marker model using enter regression

Cut-off	True positive	False negative	Sensitivity	True negative	False positive	Specificity	Overall accuracy	AUC
0.1	276	26	91.4%	465	540	46.3%	57.0%	.688
0.2	265	37	87.7%	611	394	60.1%	67.0%	.743
0.25	250	52	87.7%	693	312	60.1%	67.0%	.743
0.3	250	52	82.8%	693	312	69.0%	72.1%	.759
0.31	250	52	82.8%	693	312	69.0%	72.1%	.759
0.32	250	52	82.8%	693	312	69.0%	72.1%	.759
0.33	250	52	82.8%	693	312	69.0%	72.1%	.759
0.332	250	52	82.8%	693	312	69.0%	72.1%	.759
0.335	250	52	82.8%	693	312	69.0%	72.1%	.759
0.337	250	52	82.8%	693	312	69.0%	72.1%	.759
0.34	236	66	78.1%	720	285	71.6%	73.1%	.749
0.35	215	87	71.2%	780	225	77.6%	76.1%	.744
0.4	215	87	71.2%	780	225	77.6%	76.1%	.744

2.4.4 Cross Validation

Finally, to determine whether this model was effective outside of the modelling sample and achieve Aim 3 of the study, the weighted algorithms for aggression and destruction were calculated in the Test Data sets for cross validation. The cut-offs identified (0.27 for aggression, 0.3 for destruction) were then applied to determine the sensitivity and specificity of the algorithm within the test data set.

Table 2.13 shows that the optimal cut-off identified in the Modelling Sample was replicated in the Test Sample. The cut-off of 0.27 had a sensitivity of 80.5% and specificity of 63.8%. Overall accuracy was 72.9% with an AUC value of .719. This ensures 124 individuals known to show aggression were correctly identified, 43 were classified as aggressive that did not meet aggression criteria, 31 were considered not aggressive that were, and 87 were correctly identified as not aggressive.

Table 2.13 Cross validation of predictive power of aggression risk model

Cut-off	True positive	False negative	Sensitivity	True negative	False positive	Specificity	Overall % accuracy	AUC
0.1	144	11	93.5%	49	81	37.8%	68.0%	.653
0.2	134	21	87.0%	65	65	50.0%	70.1%	.682
0.25	128	27	83.1%	79	51	60.8%	72.9%	.717
0.26	126	29	81.8%	81	49	62.3%	72.9%	.718
0.27	124	31	80.5%	83	47	63.8%	72.9%	.719
0.28	121	34	78.6%	87	43	66.9%	73.2%	.725
0.3	118	37	76.6%	88	42	67.7%	72.5%	.719

Cross validation of the destruction risk marker model (Table 2.14) indicated a cut off of 0.3 maximised sensitivity to a sufficient level, without over-compromising specificity. This resulted in a model with 90% sensitivity and 61% specificity; 73.2% overall accuracy. This ensures 127 individuals known to show destruction were correctly identified, 64 were classified as destructive that did not meet destruction criteria, 19 were considered not destructive that were, and 100 were correctly identified as not destructive.

Table 2.14 Cross validation of predictive power of destruction risk model

Cut-off	True positive	False negative	Sensitivity	True negative	False positive	Specificity	Overall % accuracy	AUC
0.1	141	5	96.6%	43	121	26.1%	59.4%	.614
0.2	135	11	92.5%	77	87	47.0%	68.4%	.697
0.25	135	11	92.5%	77	87	47.0%	68.4%	.697
0.3	127	19	90.0%	100	64	61.0%	73.2%	.740
0.35	127	19	90.0%	100	64	61.0%	73.2%	.740
0.4	123	23	84.2%	106	58	64.6%	73.9%	.744

2.5 Discussion

The current study aimed to increase understanding of risk markers for aggressive and destructive behaviours, and translate these data into a clinical algorithm to model risk. The large dataset enabled the identification of a number of risk markers, contributing positively to the theoretical basis upon which interventions will be based and meeting the research need highlighted in Chapter 1. A particular strength of the current methodology is that it made use of a large sample that would be difficult to acquire without the support of three separate studies. This study also demonstrated that it was possible to translate these data into a statistical model of risk. The clinical algorithm developed showed good sensitivity and specificity both during modelling and cross validation. The high sensitivity showcased for both aggression and destruction algorithms suggests that, whilst this is only the preliminary stages in this area of research, it suggests it is a viable avenue to pursue to determine if it is truly possible to eventually identify children that are at greatest risk of showing aggression and destruction that may benefit from intervention.

2.5.1 Risk markers

Identification of risk markers has considerable clinical and theoretical utility. Risk markers help to identify mechanisms implicated in the development of aggressive and destructive behaviour thereby informing pathways for possible prevention of aggressive or destructive behaviours. The identification of autism and overactivity as significant risk markers for aggression is consistent with the literature base (Bamford et al., in review; Davies & Oliver, 2016). Interestingly, previous research has noted that severity of intellectual disability (termed ability in this study) is significantly associated with aggression (Bamford et al., in review); however ability was not significant in the relative risk analysis or the regression model. The present study identified that frequency of repetitive behaviour, higher impulsivity, higher overactivity, higher obsessions and presence of autism, male gender, increased health problems

and epilepsy all increase risk of aggression. Regarding destruction, the present study identified risk markers of gender, autism, impulsivity, overactivity, obsessive behaviour, health problems and repetitive behaviour. The research base regarding risk markers for destruction is considerably smaller than for aggression, however autism and repetitive behaviour have previously been indicated as variables of interest (Bamford et al., in review; Davies, & Oliver, 2016). It is interesting to note that, whilst there are similarities in the general risk markers associated with each behaviour (e.g. impulsivity, overactivity, autism, gender), the markers identified as most significant in each algorithm for aggression and destruction are different, which lends further support for the argument of studying the two behaviours separately, rather than as one conflated behaviour (Bamford et al., in review).

2.5.2 Predicting behaviour

Following the identification of the risk markers, the cut off range identified in the modelling sample for the weighted combination of these was tested in the test sample. For aggression, the results were very positive, with an AUC of .719 during cross validation, giving a sensitivity of 80.5% and specificity of 63.8%. An AUC value of one indicates an exceptionally good classifier, and for medical diagnosis, values of .95 or higher are preferred (Rice & Harris, 2005). A value of .70 and above is considered to be indicative of a strong effect for prediction of future behaviour in applied psychology (Rice & Harris, 2005). In the test sample, this cut off resulted in 43 participants being classified as likely being aggressive, despite their true score being 'not aggressive'. This would mean that 47 people in a sample of 274 would be recommended to receive therapeutic intervention that may not be necessary for them and 31 would miss out on the opportunity for intervention. The specificity of the developed algorithm is lower than would be desired; it would be considered more ideal to increase the specificity to ensure that funds are utilised in the most effective way possible. The specificity in the current study could be low for a number of reasons. One possibility is that there may be protective

factors involved that have yet to be understood in the current literature. An individual may have all the predictive covariates that would suggest they should demonstrate a particular behaviour, however their support network has been sufficiently robust to prevent this onset. Parents may have already received support to mitigate the behaviours. Similarly, it may be that an individual has all the covariates, however they have not yet started to demonstrate the expected behaviour. It is on this basis that it would be advantageous to consider a longitudinal study, to determine if the behaviours emerge later.

It is important, to consider these results in the context of the likely formats for early intervention. Such intervention strategies would most likely focus on psychoeducation and parent training, and therefore would be a supportive and likely valuable experience, even if a child is not at significant risk of developing behaviours that challenge. It is on this basis that it was deemed more beneficial to maximise sensitivity with a compromise of specificity. Once aggressive behaviour enters a child's behaviour repertoire, it is highly persistent, with the financial cost to services for adults with severe behaviour that challenges indicated as being between £89,335 and £358,415 (Emerson et al., 2014). The increased financial cost and decreased quality of life associated with these behaviours (Knapp et al., 2005) suggests that it is more important to identify as many individuals as possible to minimise the likelihood of individuals missing out on potentially beneficial interventions.

Regarding the destruction model, whilst only two risk markers were combined to predict presence of destruction (autism and impulsivity), the results showed good consistency in cross validation. The initial cut-off range posed by the modelling sample was 0.3. This cut off showed good sensitivity and specificity in the modelling sample. When this was applied to the test sample, it offered 90% sensitivity and 61% specificity. This was an overall accuracy of 73.2% and an AUC of .740. In this case, 64 participants out of 310 would have received an intervention that were not classified as showing destructive behaviour and 19 would miss out

on an intervention that may need it. Whilst it is necessary to test both of these models further in additional samples, using longitudinal designs to examine behaviour onset and changes in severity, the preliminary results for both the aggression and destruction model are promising and represent a significant advance in empirical research in this area, which has traditionally only described associations between risk markers and behaviours than challenge.

2.5.3 Clinical utility

This study has demonstrated that there is potential to be pursued regarding identifying individuals who are at greatest risk of aggressive and destructive behaviour. The SAD-SQ(R) is a brief questionnaire that participants were able to complete alone in a short space of time. This, therefore, represents a valuable tool for clinicians and a continuation of the current research to determine the predictive utility of this measure to identify 'at risk' individuals, rather than individuals with currently present behaviour, would be beneficial. The results also suggest that an early intervention pathway, similar to that seen in psychosis (where young people are identified as 'Ultra High Risk' for psychosis), may be viable for people with intellectual disabilities to ameliorate aggression and destructive behaviour.

The need for prevention and early recognition of behaviours that challenge has long been recognised in this field, however Chapter 1 emphasised that there is a lack of understanding of the risk markers that may enable early identification in the future. The current study has commenced the first step in determining the feasibility of early identification and shows promising prediction of currently present behaviour, suggesting it is a positive avenue to pursue.

In order for an intervention to be devised, it is necessary to have a cost-effective, efficient and relatively reliable strategy to identify individuals at risk. The cut-offs selected were chosen to ensure that valuable funds were not used to provide every individual with ID with an

intervention; rather it has demonstrated that it is possible to include as many individuals as possible without being over-inclusive. The impact of early intervention in other areas, such as psychosis, has been immensely positive, with Tsiachristas, Thomas, Lean and Lennox (2016) demonstrating that individuals in early intervention for psychosis services were 116% more likely to go on to be employed and 52% more likely to live in mainstream accommodation. Moreover, ratings of emotional well-being were 17% more likely to improve. In terms of finance, Tsiachristas et al. (2016) estimated that the saving in NHS costs would be £33.5 million per annum, and societal cost savings could reach £63.3 million per annum. The financial relief that early intervention for aggressive and destructive behaviour has the potential to bring to the healthcare system is substantial, and therefore this represents an avenue with great potential.

2.5.4 Additional considerations

The current study is not without limitation. Statistical analysis between the Modelling and Test samples indicated that there were significant differences in terms of prevalence of aggression and destruction. The prevalence of aggression in the Modelling sample was 30.2%. This suggests that aggression was not highly frequent and the model designed had to be relatively accurate to correctly classify individuals. For the Test sample, however, prevalence was 54.2%. This is essentially half of the sample, which increases the likelihood of individuals being correctly classified based on chance, not an accurate model. With this said, however, the Test Sample was recruited from a population specifically chosen as it represents the population most likely to be chosen for early intervention (young children) and also from locations where screening is more likely to take place (Child Development Centres, clinics and special schools). It is therefore most essential that the model is accurate within this population, however further analysis in similar samples will be beneficial to determine the consistency of the sensitivity and specificity of the identified model.

A further consideration is that within the Destruction Modelling Sample, a decision was made to include speech and ability deficits, despite this inclusion resulting in a smaller than ideal sample size and likely increasing the risk of Type 2 error. It is therefore possible that risk markers that were considered insignificant and excluded from the model may have been subject to a Type 2 error and additional research with larger sample sizes should be conducted using all variables before concluding that presence of autism and impulsivity are the most predictive markers. With this being said, the destruction model based solely on these two markers was effective in identifying individuals that currently show destructive behaviour, suggesting that they are robust markers.

There could be an alternative argument for the method of deciding whether aggression or destruction was present in the samples; a score of 1, 2, 3 or 4 for frequency of behaviour all suggest that the behaviour has, at some point, occurred. A weakness of the SAD-SQ model, however, is that a score of 1 does not provide any additional space for the participant to elaborate on when the behaviour occurred or to what extent it would induce clinical concern. In order to ensure that the algorithm was based on identifying individuals that are likely to present at some point in clinics to request support for behaviour, the clinical decision was made to only consider scores of 1 or 2 if the parent was particularly concerned about the behaviour (scoring a 3 or 4 for concern, and therefore suggesting they would present for support). It is believed that the decision to categorise behaviour in this way is a strength of the research as it would promote increased specificity from the algorithm.

2.5.5 Conclusion

In summary, this study positively contributes to the deficit in research and knowledge regarding risk markers for aggressive and destructive behaviours. It also indicates that there is realistic potential to develop a quick, effective and reliable algorithm, based on stable risk markers, to identify individuals most at risk of developing aggressive and destructive behaviour. The

ability to identify current behaviour based on predictor risk markers could open up the possibility of an early-intervention approach for aggression and destruction, which has the potential to improve the quality of life of individuals with intellectual disabilities and alleviate the financial stress that such behaviours place on the healthcare system in the long-term. The next steps in research in this field will be looking at whether children can be classified as 'at risk' at an early age based on the risk markers, and then watch their progression to see if this prediction becomes accurate.

CHAPTER 3
Executive Summary

3.1 Meta-analysis

3.1.1 Background

Intellectual disability (ID) is defined as a chronic, severe condition that limits an individual's adaptive behaviour and intellectual functioning (World Health Organisation, 1992). Approximately one in 100 individuals with ID will show behaviour that is considered challenging at some point during their lives (Emerson et al., 2001) and this is unlikely to go away if it is not treated (Totsika & Hastings, 2009). Self-injury, aggression and destruction have been a particular focus of research and the presence of behaviours is associated with psychiatric hospitalisation, reactive physical intervention and reduced quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009; Mandell, 2008; Allen, Lower, Brophy & Moore, 2009). The term 'self-injury' or 'self-injurious behaviour' (SIB) includes behaviours that are self-directed and cause physical harm to the individual, such as biting self, pulling own hair or head banging (Rojahn et al., 2008). Research has predicted that between 4 in 100 and 24 in 100 people with ID will engage in this behaviour (Cooper et al., 2009; Deb, Thomas & Bright, 2001). 'Aggression' can be used to refer to an array of acts, including physical assaults on peers, family members or staff, hostility or verbal threats (Rojahn et al., 2001). Research predicts between 30 in 100 and 60 in 100 people with ID will do this (Crocker et al., 2006; Cohen et al., 2010). To date, research has often considered 'destruction' under the umbrella of aggression, making it difficult to isolate the two forms of behaviour and consider risk markers associated with destruction specifically. A previous meta-analysis by McClintock, Hall and Oliver (2003) summarised risk data for SIB, aggression and destruction of property prior to 2002. They were able to look at the following risk markers for each behaviour: SIB (gender, degree of ID, autism, receptive communication and expressive communication), aggression (gender, degree of ID, autism and expressive communication) and destruction of property

(autism). The present meta-analysis aimed to update this research and summarise data on demographic, diagnostic and health/ person/behavioural risk markers for SIB, aggression and destruction in people with ID.

3.1.2 What did the study do?

The study conducted a large literature search and identified a total of 60 papers that provided enough information for statistical tests. This involved, for example, the number of males that showed aggression and the number that didn't, to determine the risk of gender, in this example. It did this for all of the risk markers that could be identified (21 in total) for each of the three behaviours.

3.1.3 What did the meta-analysis find?

This meta-analysis found that there was a lot more research focussing on risk markers for self-injury than there was for aggression and destruction. This enabled a good understanding of risk markers for self-injury, however it is clear that more research needs to be done in the fields of aggression and destruction. The summary of the risk markers that were significant for each behaviour is presented in Figure 3.1.

3.1.4 What does this mean?

This meta-analysis shows that the research into risk markers for these behaviours has grown considerably in the past 15 years since the previous summary was conducted. This is incredibly useful to know, as understanding factors about people that increase their risk of particular behaviours can help us firstly to identify them as early as possible, and secondly to start looking at developing interventions that are informed by these risk markers, to ensure interventions are as good as they can be in this population. If it were possible to use these risk markers to identify people earlier, possibly before behaviours develop, it might be possible to save the NHS a lot of money.

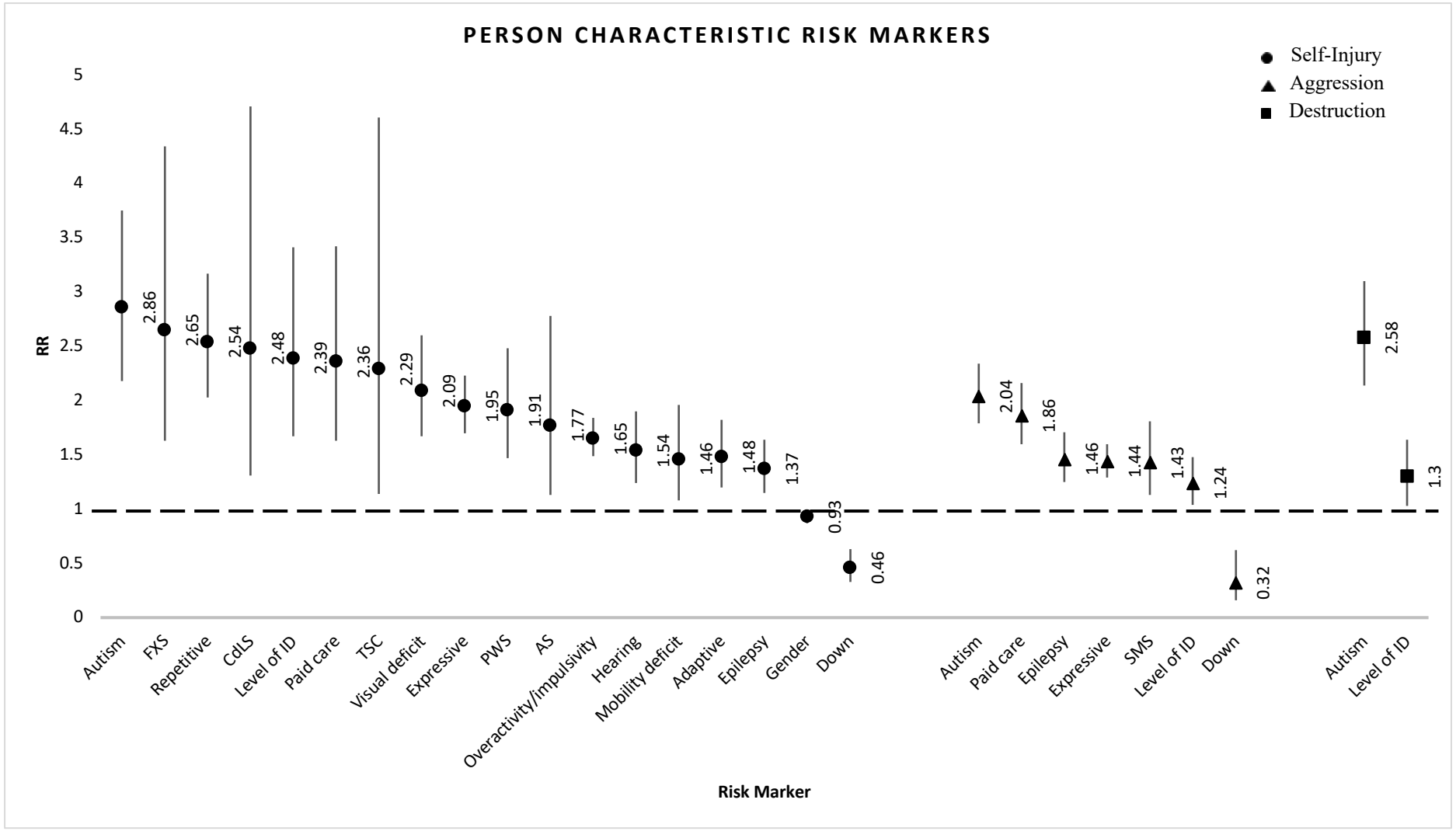

Figure 3.1 Summary of all risk markers that significantly associated with each behaviour studied.

3.2 Empirical paper

3.2.1 Background

Of the proportion of typically developing children that engage in some form of aggressive behaviour (including destruction), only 5-10% are likely to continue this behaviour into adulthood (Moffitt, 1993; Tremblay et al., 2004) and only 5% of those with extreme behaviour in childhood are likely to experience adverse impacts, such as drug addiction and criminality by age 25 (Fergusson et al., 2005). In contrast, individuals with ID that engage in behaviours that challenge are more likely to experience reactive physical intervention, psychiatric hospital intervention, school exclusion and decreased quality of life (Beadle-Brown, Murphy & DiTerlizzi, 2009; Mandell, 2008; Knapp et al., 2005). One important limitation of current interventions is that they typically depend on the behaviour already being present and the individual coming into contact with appropriate specialised, high-cost services (Feldman et al., 2004) and successful interventions often necessitate highly controlled environments and precise management.

Early intervention is characterised as an attempt to delay or prevent problems from developing by intervening before the problem emerges, based on factors about a person that suggest they are at risk (Ramey & Ramey, 1998). Early intervention has proven effective for treatment of psychosis, with reduced chances of those at-risk developing psychosis (McGorry et al., 2002). Moreover, early psychosis services are often more cost effective than standard services as there is less need for costly inpatient care (McCrone et al., 2009). Therefore, the current study sought to explore if this was an option for aggression and destruction in children with ID.

3.2.2 What did the study do?

In order to see if this route is possible, the study used two different samples of participants and had them complete a questionnaire about different known risk markers (age, gender, presence

of autism, among others). It also asked for the frequency at which aggression and destruction currently occur. This information was then used to statistically determine which risk markers were most predictive of the current presence or absence of aggression and destruction. Once these risk markers were established, the second data set was used to see if they were as good at predicting people that currently showed aggression and destruction in a different data set. The study needed to make correct identification of those with the behaviour as high as possible, whilst still maintaining a good level of accuracy of determining those that don't (rather than just saying everyone has it). This is because future models would look to be used as the basis of an intervention – cost effectiveness requires this to be not over-inclusive, but also to be good at detecting people that would benefit from an intervention.

3.2.3 What did the study find?

The markers that were identified for as most effective in predicting aggression when combined together were: gender, presence of autism, skin problems, number of health problems, repetitive behaviour, obsessive behaviour and impulsivity. When these risk markers were applied to the second data set, they identified 80.5% (124 people) that were aggressive correctly and 63.8% (83 people) that were not correctly. 31 were considered not aggressive that were, and 47 were predicted to be aggressive that weren't. For destruction, presence of autism and impulsivity were the only two risk markers that predicted presence of destruction well. In the second sample, this combination correctly identified 90% of those that were destructive correctly (127 people) and 61% of those that were not correctly (100 people). 64 people were classified as destructive that did not meet destruction criteria and 19 were considered not destructive that were.

3.2.4 What does this mean?

This result suggests that there may be some benefit in continuing to think about early intervention as a strategy for aggression and destruction in ID. The model was good at detecting

people with the behaviour, and not too many people were either included that should not have been, or were excluded that shouldn't have been. This model is only a starting point, as it is looking at behaviour that is currently present. The next steps in research in this field will be looking at whether children can be classified as 'at risk' at an early age based on the risk markers, and then watch their progression to see if this prediction becomes accurate.

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Appendix A: Descriptions of studies included in meta-analysis

Reference	Behaviour	Measure	Risk Markers	Country	Sample description
***Ando & Yoshimura (1979a, b)**	Self-injury; Aggression; Destruction	Maladaptive behaviour checklist (Aman, 1994)	Receptive/ expressive communication; ASD; Psychosis	Japan	47 autistic children and 128 children with intellectual disabilities (age 6-14)
Arron et al. (2010)	Self-injury; Aggression	Challenging Behaviour Questionnaire (Hyman et al., 2002)	AS; CdLS; Adaptive function; CdCS; FXS; PWS; Lowe syndrome; SMS	USA	741 participants recruited through specific syndrome societies, foundations and support groups.
***Ballinger (1971)**	Self-injury:	Self-determined measure	Gender; Degree of ID; Psychosis; Psychotropic medication	Scotland (UK)	626 adult residents in a hospital/institution for 'mentally subnormal'
Barnard-Brak et al. (2014)	Self-injury	Behaviour Problem Inventory (Rojan et al., 2001)	Gender; Age; Degree of ID	International	1871 cases from five regions around the world
Basile et al. (2007)	Self-injury	Developmental Behaviour Checklist (Einfeld & Tonge (1992)	Adaptive function	Italy	56 individuals with CdLS. Recruited through the Italian Cornelia de Langue Association
***Bhaumik et al. (1997)**	Self-injury; Aggression, Destruction	Self-determined measure	Autism	UK	2261 adults with intellectual disabilities served by a regional intellectual disability service
***Bott et al. (1997)**	Self-injury; Destruction	Self-determined measure	Expressive communication	UK	Individuals in 13 health districts' listed Registers for People with Intellectual Disabilities
Bowring et al. (2017)	Self-injury	Behaviour Problem Inventory (Rojan et al., 2001)	Degree of ID; Gender; ASD; DS; Incontinence; Visual impairment; Mobility impairment; Hearing impairment; Seizures; Dementia; Age; Receptive/ expressive communication; Epilepsy; Living arrangement	Jersey	265 adults known to services and administratively defined as having ID.
Cohen et al. (2010)	Aggression	Modified Overt Aggression Scale (Kay et al., 1988)	Age; Gender; Level of ID; ASD	USA	3547 individuals with ID and/or autism. 897 comprising an aggression sample.
Cooper et al. (2009)	Aggression	Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation	Gender; Degree of ID; Expressive communication; ASD; ADHD; DS; Epilepsy; Visual impairment; Mobility impairment; Hearing impairment; Living status; Age; Incontinence	Scotland, UK	All adults with ID within a geographically defined area of Scotland, UK

Crocker et al. (2006)	Self-injury; Aggression; Destruction	Modified Overt Aggression Scale (Kay et al., 1988)	Level of ID; Age; Living status	Quebec, Canada	1365 adult men and women receiving input from rehabilitation agencies
Crocker et al. (2007)	Self-mutilation	Modified Overt Aggression Scale (Kay et al., 1988)	Gender; Degree of ID; Mobility impairment; Living status; Age; Overactivity (Reiss Screen – not included)	Quebec, Canada	296 adults with mild/moderate ID interviewed and client files reviewed
***Davidson et al. (1994)**	Aggression		Degree of ID; ASD; Gender	UK	199 individuals referred to a crisis intervention program
Davies & Oliver (2016)	Self-injury; Aggression; Destruction	SAD-SQ (Davies & Oliver, 2016)	Gender; Degree of ID; Overactivity and impulsivity (SAD-SQ); Repetitive and restricted behaviours (SAD-SQ)	UK	417 children with severe intellectual disability
Deb et al. (2001)	Self-injury; Aggression	Disability Assessment Schedule (Holmes et al., 1982)	Gender; Age; Living status; Degree of ID; Epilepsy; Expressive communication; Mobility impairment	Wales, UK	One hundred and one adults with ID aged between 16 and 64 known to the Vale of Glamorgan Social Services Department in South Wales, UK.
Eden et al. (2014)	Self-injury; Aggression	Challenging Behaviour Questionnaire (Hyman et al., 2002)	TSC; CdLS; FXS; ASD; DS; Age (not included); Gender; Interest/pleasure; Socialisation; Repetitive behaviour; Impulsivity/ compulsivity; Overactivity; Stereotyped behaviour; Pain; Health conditions	UK	37 children aged 4 to 15 years, with TSC.
Emmerson et al. (2001)	Self-injury	Unspecified interview schedule	ASD; Age; Expressive/ receptive communication	UK	95 men and women (age 12-65)
***Eyman & Call (1977)**	Self-injury; Aggression		Degree of ID	UK	6870 individuals from institutions and community centres
Folch et al. (2018)	Self-injury	Aberrant Behaviour Checklist (Aman & Singh, 1994)	Age; Gender; Degree of ID; Place of residence; Oral pain; Epilepsy; CARS scores; Psychiatric medication	Spain	953 individuals with ID recruited from 66 social care services for ID.
***Griffin et al., (1986)**	Self-injury		Gender		Residents of all 13 state institutions
***Hardan & Sahl (1997)**	Self-injury Aggression	Unspecified	Degree of ID		Children and adolescents taking part in 12-month program for intellectual disorders
Hemmings et al. (2006)	Self-injury; Aggression; Destruction	Disability Assessment Schedule (Holmes et al., 1982)	Age; Degree of ID; Energy; Anhedonia; Mood; Social impairment; Sleep; Appetite	UK	739 adults aged 18-85 known to a local register for people with ID in Southeast London

Hiraiwa et al. (2007)	Self-injury	Unspecified questionnaire	Gender, Age; BMI; Degree of ID	Japan	177 participants with PWS recruited through connections with Japanese PWS Association called Takenoko-no-kai
Holden et al. (2003)	Self-injury	Observation	Degree of ID	Norway	165 adults with 'mental retardation' receiving services from rehabilitation services in Oppland and Hedmark
***Jacobson (1982)**	Self-injury; Aggression	Unspecified	Degree of ID		30,578 developmentally disabled individuals living in community and institutions in New York
***Kebbon & WIndahl (1986)**	Self-injury	Unspecified	Degree of ID		All individuals in Sweden served by services for mental retardation
***Kiernan & Alborz (1996)**	Self-injury	Unspecified	Receptive communication		Individuals from seven health districts with challenging behaviour
Langthorne &McGill (2011)	Self-Injury Aggression Destruction	Aberrant Behaviour Checklist (Aman & Singh, 1994)	FXS; SMS	UK	Individuals with ID aged 5-21 attending parental support groups for specific syndromes
Lundgvist (2013)	Self-injury	Behaviour Problem Inventory (Rojan et al., 2001)	Gender; Age; Degree of ID; Physical health difficulties; Epilepsy; Cerebral Palsy; PWS; DS; FXS; ASD; ADHD; Medication; Communication; Hypersensitivity; Dementia; Psychosis; Depression;	Sweden	915 individuals over the age of 18 receiving at least minimum care from local health authorities.
***Maisto et al. (1978)**	Self-injury	Unspecified questionnaire	Degree of ID; Gender	USA	1,300 residents in a large residential training institution in US
Matson et al. (2008)	Self Injury; Aggression; Destruction	Autism Spectrum Disorders – Behaviour Problems for Adults.	ASD	USA	320 adult residents from two developmental centres in Louisiana.
***Maurice & Trudel (1982)**	Self-injury		Gender		2,858 individuals with intellectual disabilities living in three institutions
***McLean et al. (1996)**	Self-injury Aggression		Expressive communication		Total population of individuals with mental retardation in state of Kansas
Medeiros et al. (2013)	Self-injury	Behaviour Problems Inventory (Rojan et al., 2001)	Age, Gender, Ethnicity	USA	115 adults with varying levels of ID engaged in a day training and rehabilitation placement in Minnesota.
Newman et al. (2015)	Self-injury	Behaviour Problems Inventory (Rojan et al., 2001)	FXS	Ireland	47 children and adolescents with a diagnosis of FRX recruited through online forums and groups

Oliver et al. (2009)	Self-injury Aggression Destruction	Challenging Behaviour Interview (Hyman et al., 2002)	CdLS	UK and Ireland	54 people with CdLS and 46 people with intellectual disabilities recruited through the CdLS Parent foundation group and local area
Oliver et al. (2012)	Self-injury Aggression Destruction	5 items taken from the original Wessex scale.	Stereotyped behaviour, Communication, Adaptive behaviour	UK	1096 children attending 17 special schools for children with severe intellectual disabilities
***Quine (1986)**	Self-injury Aggression	Unclear	Gender	UK	Individuals from two health districts with intellectual disabilities
Richards et al. (2012)	Self-injury	Aberrant Behaviour Checklist (Aman & Singh, 1994)	ASD, DS, FXS, Gender, Age, Ability, Mood, Interest/pleasure, Stereotyped behaviours, Overactivity/impulsivity	UK	321 participants with ASD, Fragile X and Down syndrome recruited via National Autistic Society, Fragile X Society and Down Syndrome Association.
Richards et al. (2017)	Self-injury	SAD-SQ; Challenging Behaviour Questionnaire (Davies & Oliver, 2016)	Gender, Age, Physical health difficulties, overactivity/impulsivity, Ability	UK	515 individuals with ASD attending National Autism Society adult services and schools
***Rojahn (1986)**	Self-injury	Six-page survey instrument – SIB section adapted from Schroeder et al. (1978) and Rojan (1984).	Degree of ID	Germany	25,872 'mentally retarded' individuals using schools/training centres, workshops or group homes in Germany.
***Ross (1972)**	Self-destructive behaviour Attacks others		Degree of ID		11,139 individuals with mental retardation living in state hospitals in California in 1970
***Schroeder et al. (1978)**	Self-injury	Interview with social worker	Degree of ID; Receptive/expressive communication	USA	1,150 residents of Murdoch Centre, a state facility for 'retarded persons', aged 5-85 years.
Sheth et al. (2015)	Self-injury Physical aggression Destruction	Challenging Behaviour Questionnaire (Hyman et al., 2002)	Sotos syndrome; PWS; DS, ASD	UK	150 Participants with Sotos syndrome, Prader Willi syndrome, Autism or Down syndrome
***Shodell & Reiter (1968)**	Self-mutilation		Expressive communication		58 schizophrenic children attending daytime special education programmes
Tenneij et al. (2009)	Aggression	Staff Observation Aggression Scale – Revised (Nijman & Palmstierna, 2002); Adult	Age, Gender, Treatment duration,	Netherlands	108 adults recruited from four inpatient treatment facilities for adults with mild ID

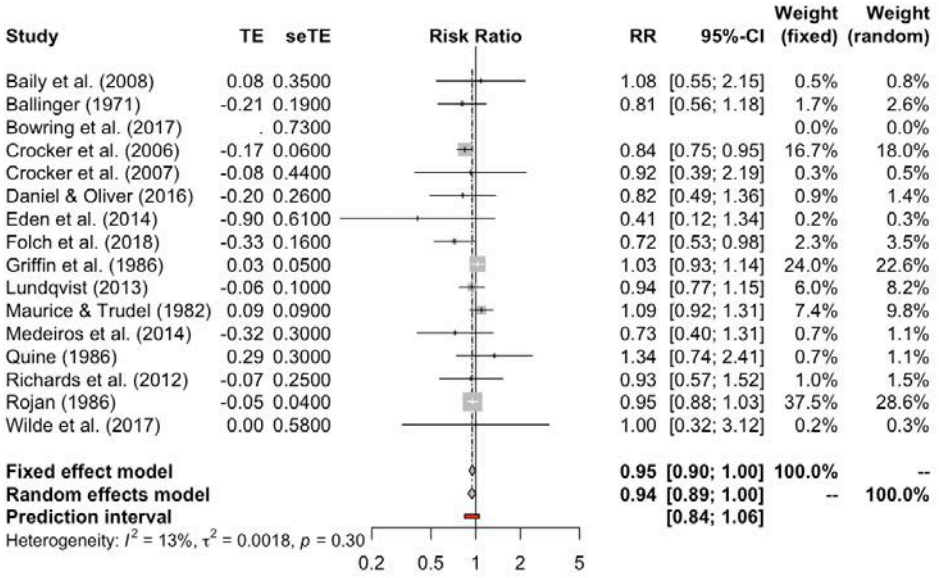
		Behaviour Checklist (Achenback & Rescorla, 2003)			and severe behavioural and emotional problems
Tsiouris et al. (2011)	Aggression	Modified Overt Aggression Scale (Kay et al., 1988)	Psychiatric disorder	USA	Individuals with ID living in the community and receiving services from the New York State Office for People with Developmental Disabilities between 2006-2007.
Tyrer et al. (2005)	Aggression	Disability Assessment Schedule (Holmes et al., 1982)	Gender, Age, Ethnicity, Living status, Degree of ID, Epilepsy, DS, ASD	UK	Cross sectional study of 3062 participants using the Leicestershire LD Register for adults over the age of 19 with LD.
Wilde et al. (2017)	Self-injury Aggression	Challenging Behaviour Questionnaire (Hyman, Oliver & Hall, 2002)	TSC, Age, Gender, Communication, Socialisation, Impulsivity, AS	UK	30 individuals with TSC recruited through Tuberous Sclerosis Association, matched to 21 individuals with DS and 29 people with ASD

*Indicates studies included in McClintock, Hall & Oliver (2003)

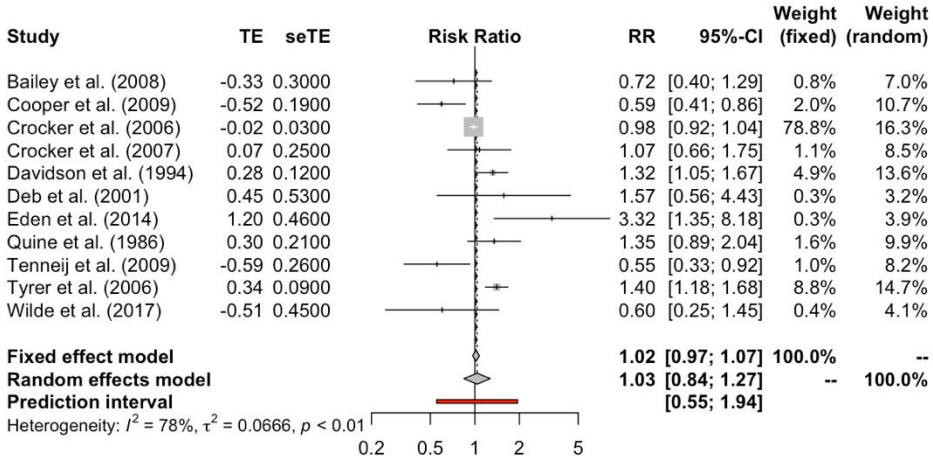
Appendix B: Forest plots of individual risk marker outputs

B1 Gender

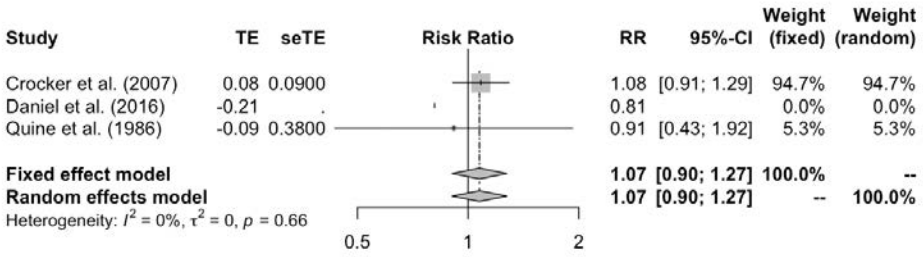
B1.1 Forest plot for all studies included for RR of gender and self-injury



B1.2 Forest plot for all studies included for RR of gender and aggression

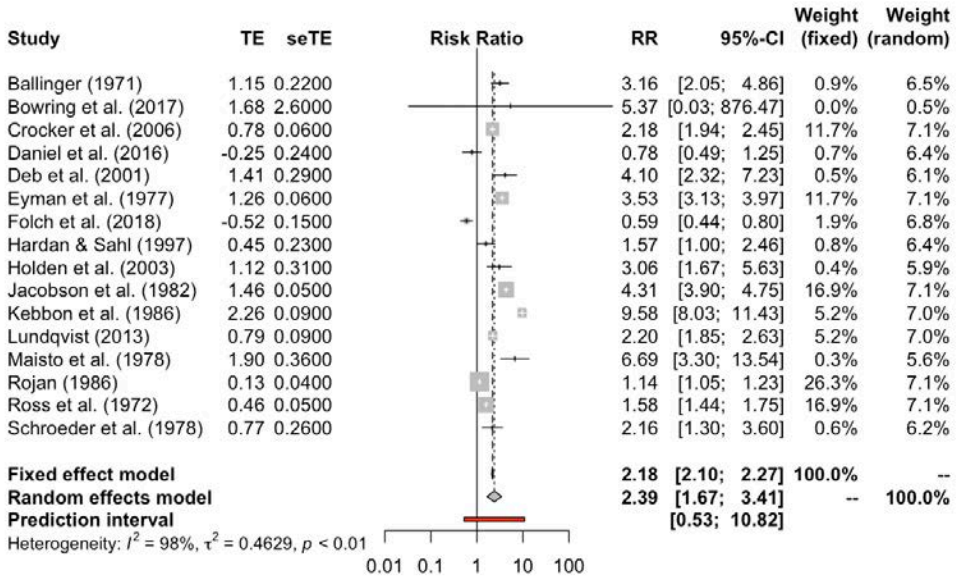


B1.3 Forest plot for all studies included for RR of gender and destruction

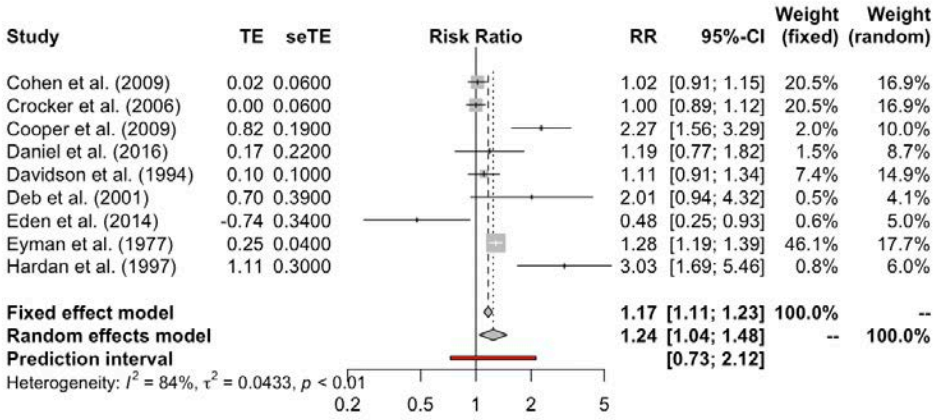


B2 Level of ID

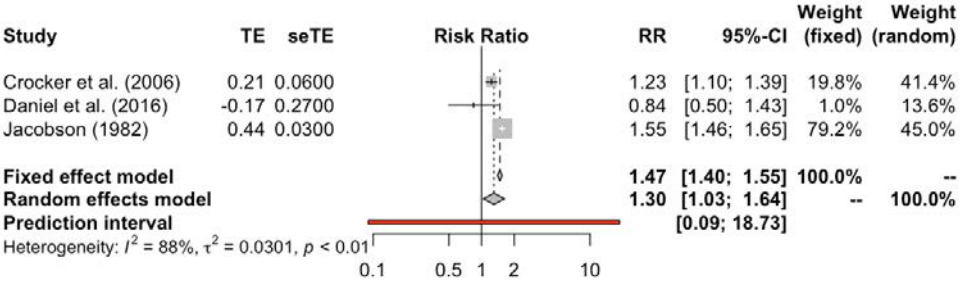
B2.1 Forest plot for all studies included for RR of level of ID and self-injury



B2.2 Forest plot for all studies included for RR of level of ID and aggression

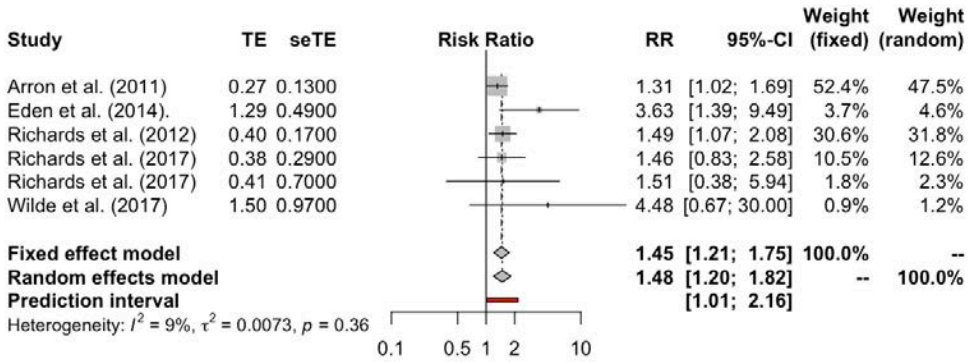


B2.3 Forest plot for all studies included for RR of level of ID and destruction

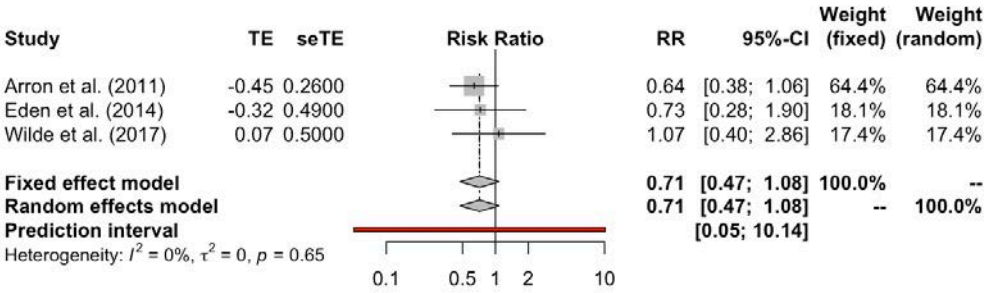


B3 Adaptive Behaviour

B3.1 Forest plot for all studies included for RR of adaptive behaviour and self-injury

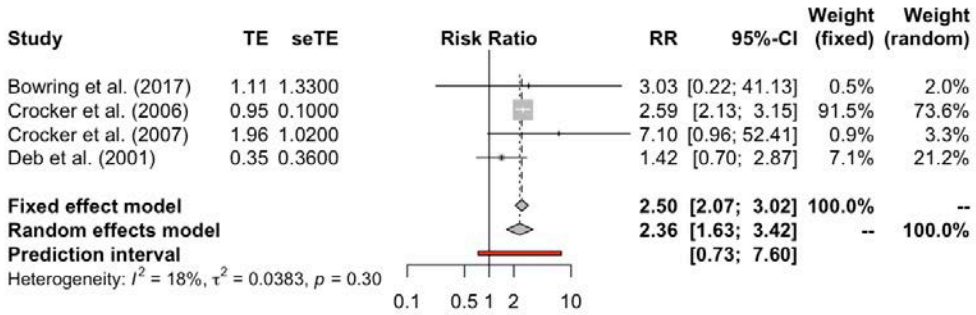


B3.2 Forest plot for all studies included for RR of adaptive behaviour and destruction

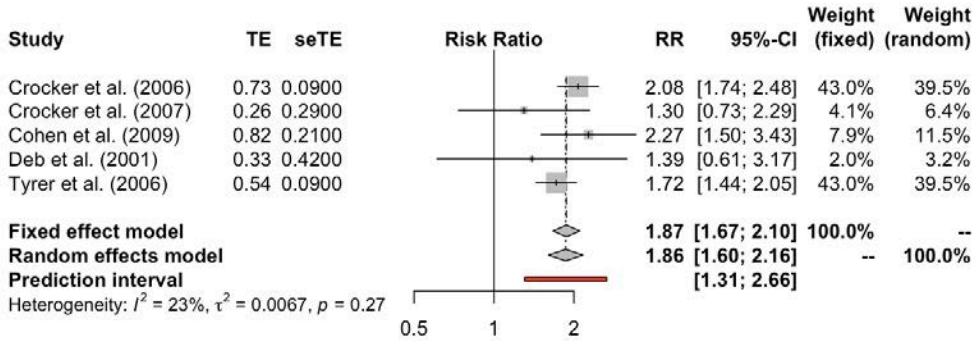


B4 Paid/Congregate care

B4.1 Forest plot for all studies included for RR of paid/congregate care and self-injury

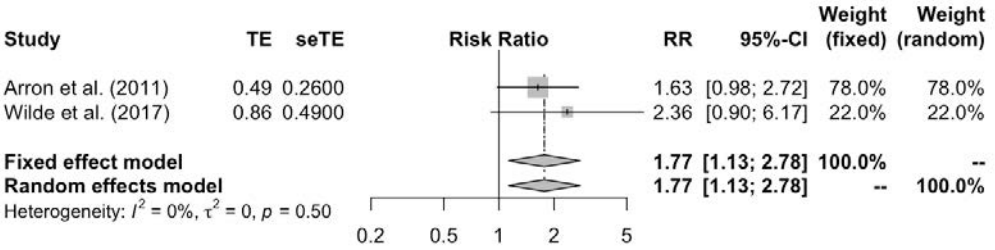


B4.2 Forest plot for all studies included for RR of paid/congregate care and aggression

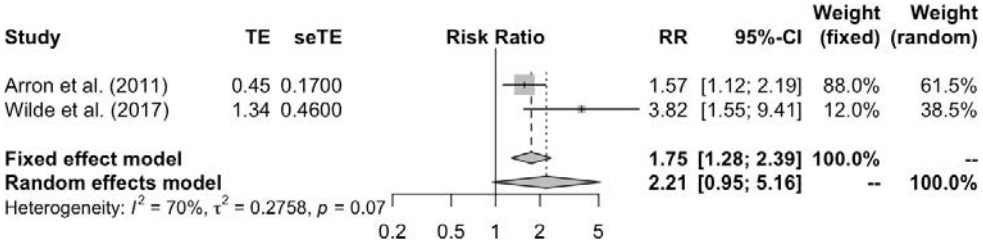


B5 Angelman syndrome

B5.1 Forest plot for all studies included for RR of Angelman syndrome and self-injury

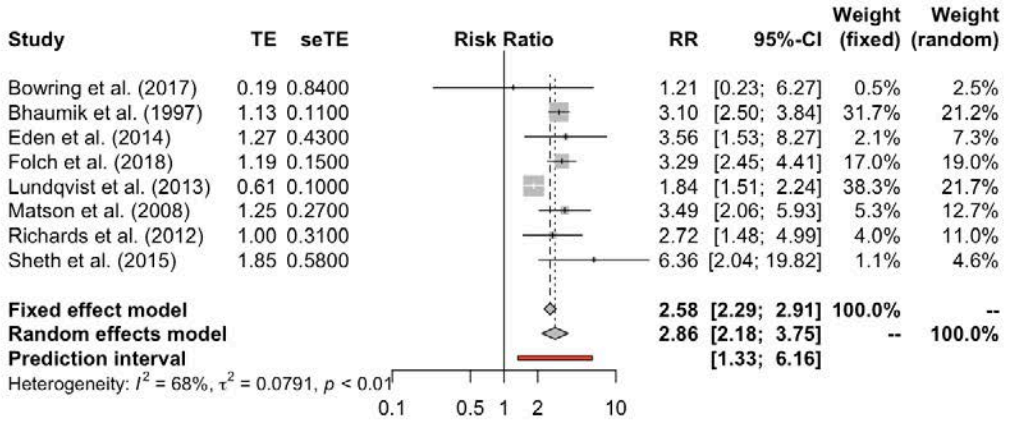


B5.2 Forest plot for all studies included for RR of Angelman syndrome and aggression

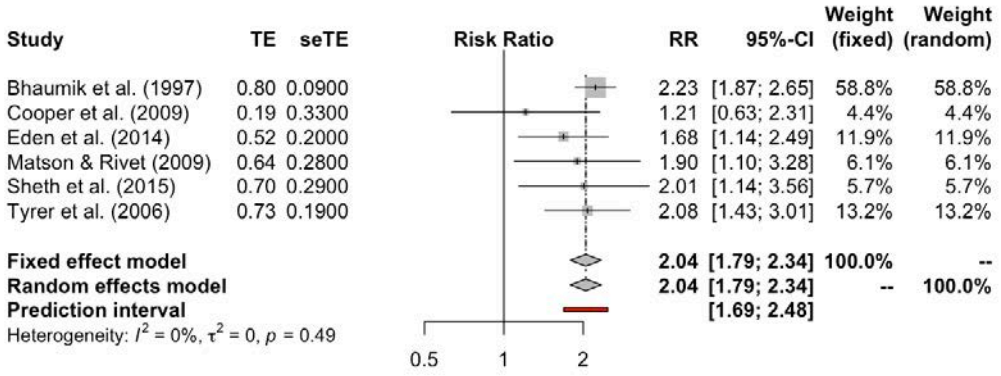


B6 Autism

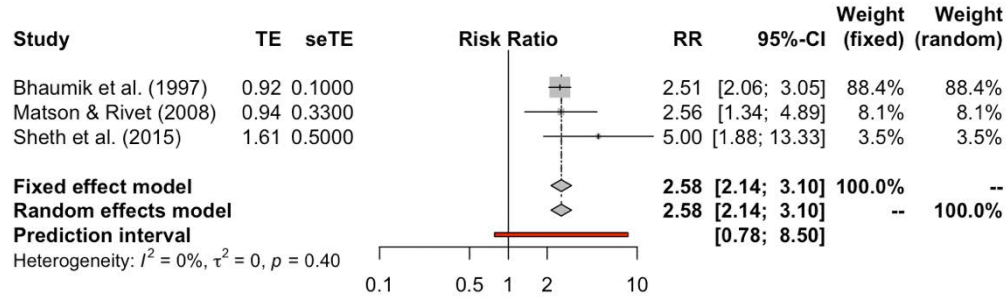
B6.1 Forest plot for all studies included for RR of autism and self-injury



B6.2 Forest plot for all studies included for RR of autism and aggression

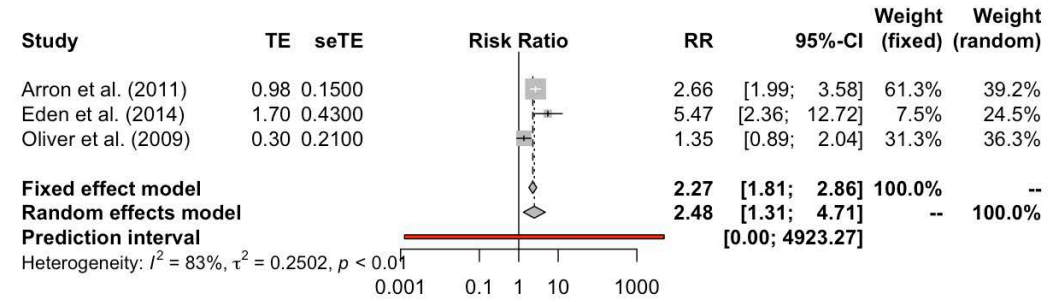


B6.3 Forest plot for all studies included for RR of autism and destruction

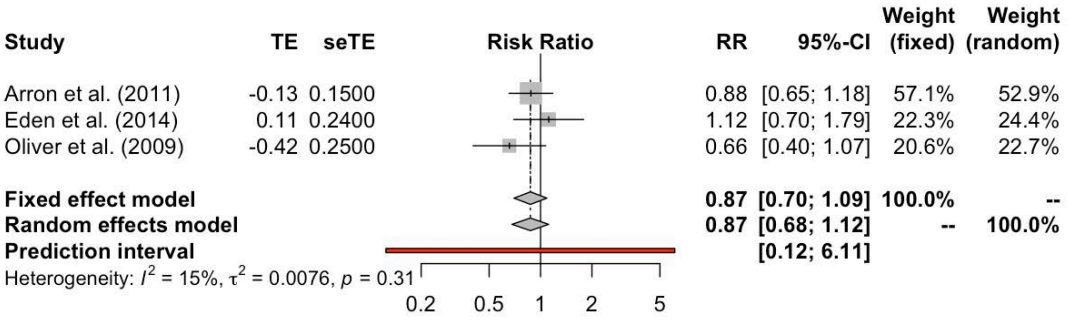


B7 Cornelia de Lange syndrome

B7.1 Forest plot for all studies included for RR of Cornelia de Lange syndrome and self-injury

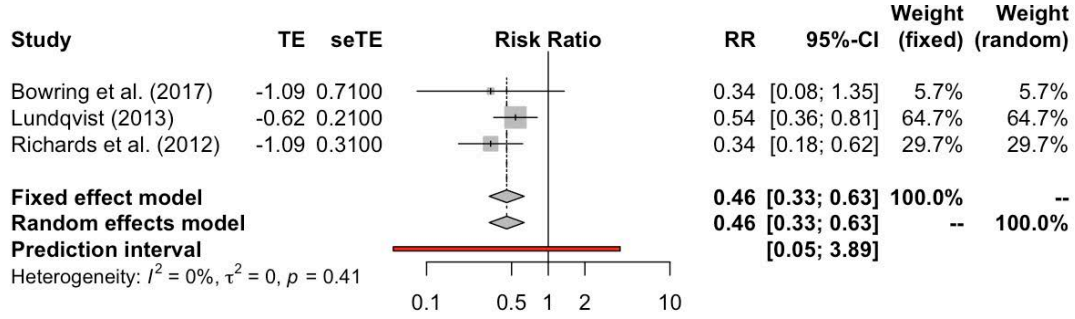


B7.2 Forest plot for all studies included for RR of Cornelia de Lange syndrome and aggression

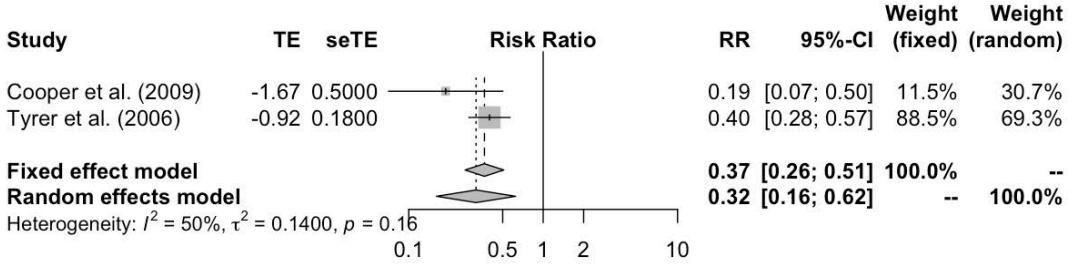


B8 Down syndrome

B8.1 Forest plot for all studies included for RR of Down syndrome and self-injury

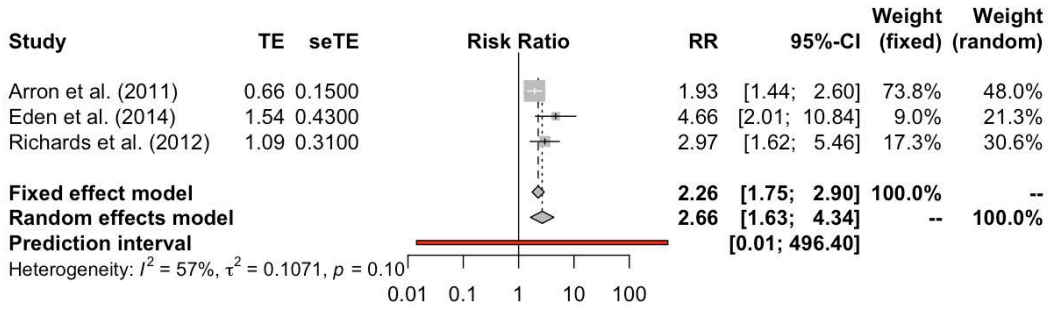


B8.2 Forest plot for all studies included for RR of Down syndrome and aggression

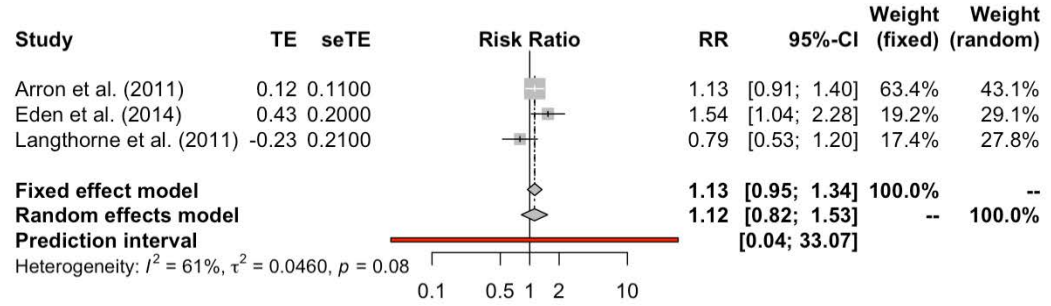


B9 Fragile X syndrome

B9.1 Forest plot for all studies included for RR of fragile X syndrome and self-injury

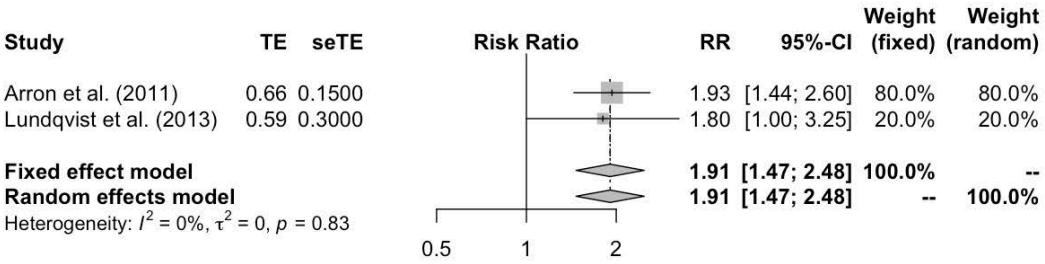


B9.2 Forest plot for all studies included for RR of fragile X syndrome and aggression

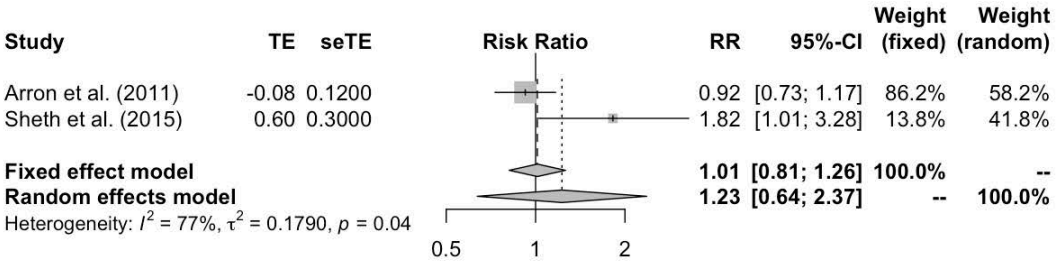


B10 Prader Willi syndrome

B10.1 Forest plot for all studies included for RR of Prader Willi syndrome and self-injury



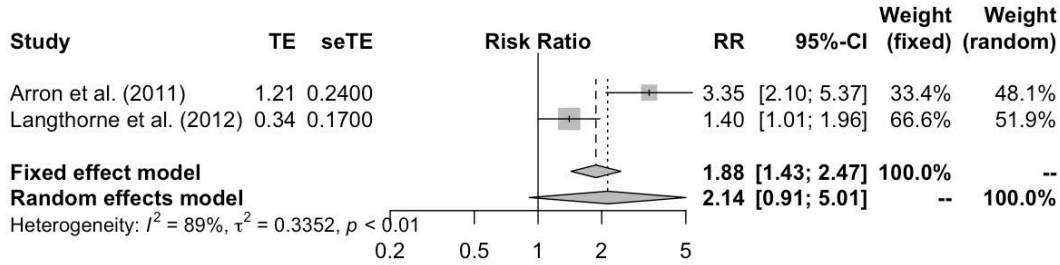
B10.2 Forest plot for all studies included for RR of Prader Willi syndrome and aggression



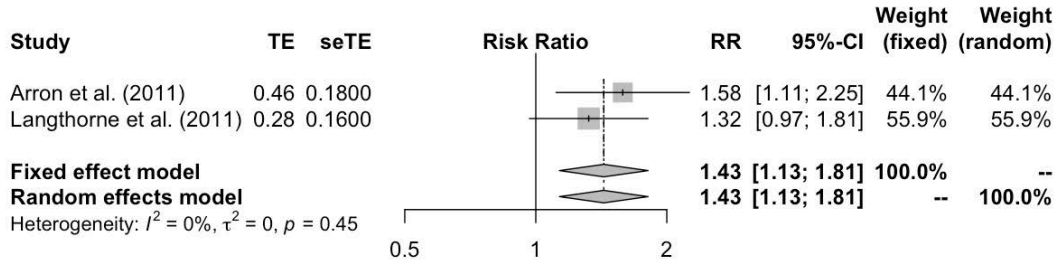
B11 Smith

Magenis syndrome

B11.1 Forest plot for all studies included for RR of Smith Magenis syndrome and self-injury

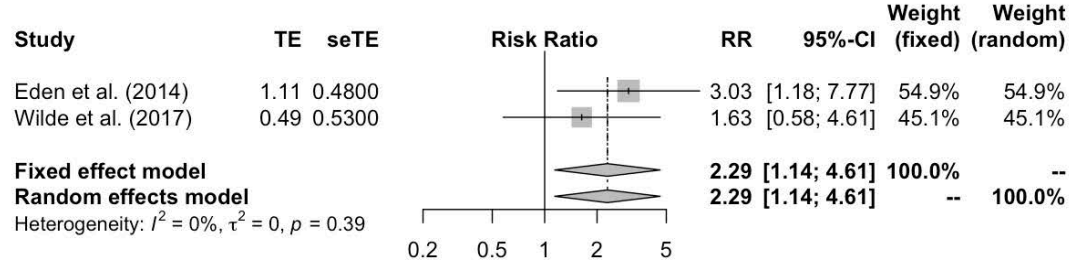


B11.2 Forest plot for all studies included for RR of Smith Magenis syndrome and aggression

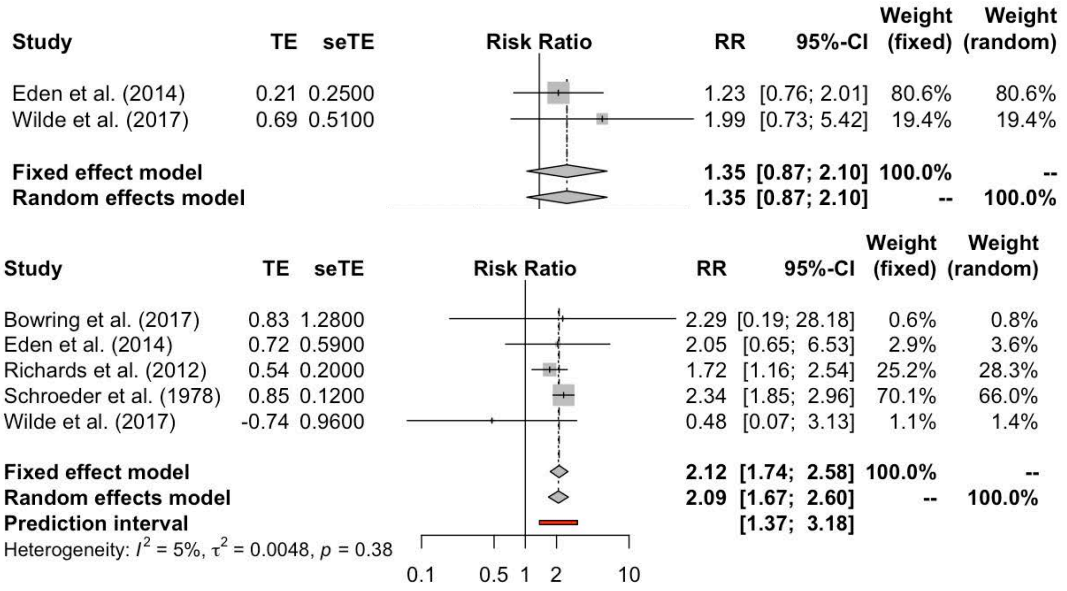


B12 Tuberous sclerosis complex

B12.1 Forest plot for all studies included for RR of tuberous sclerosis complex and self-injury



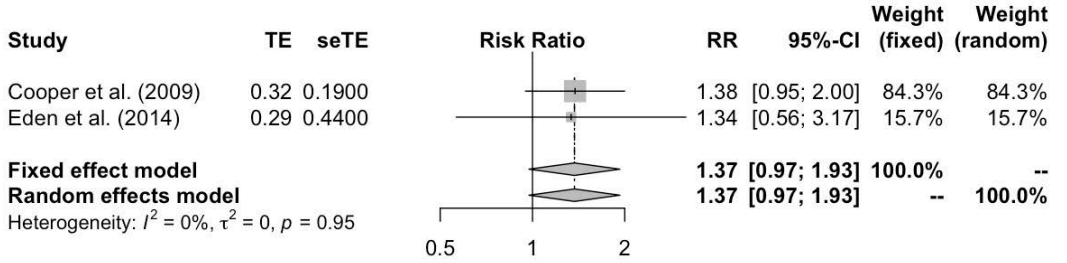
B12.2 Forest plot for all studies included for RR of tuberous sclerosis complex and aggression



B13 Visual deficit

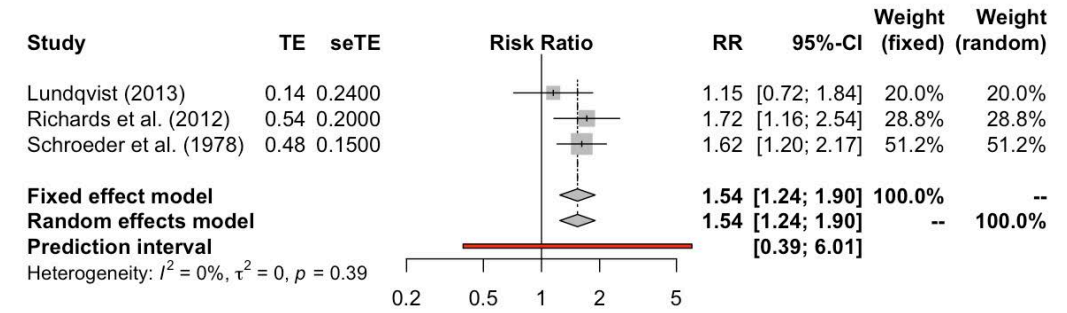
B13.1 Forest plot for all studies included for RR of visual deficit and self-injury

B13.2 Forest plot for all studies included for RR of visual deficit and aggression



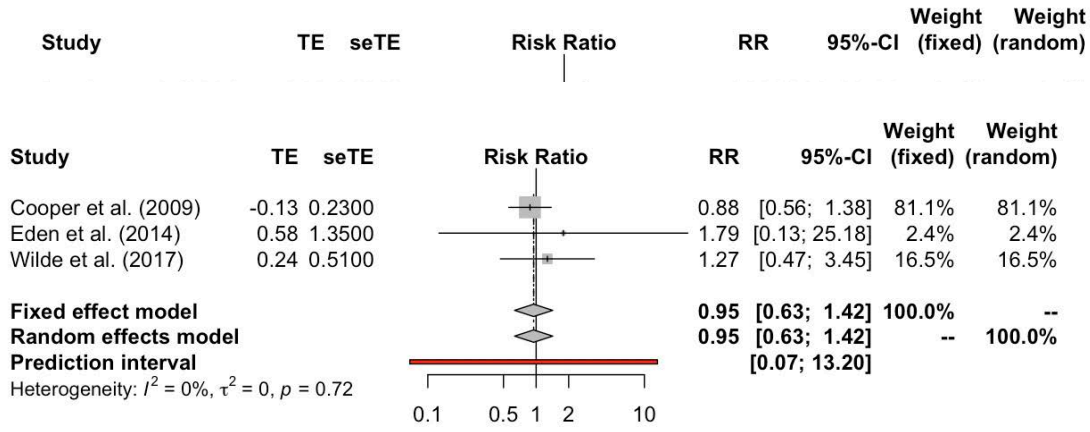
B14 Hearing deficit

B14.1 Forest plot for all studies included for RR of hearing deficit and self-injury



B15 Mobility deficit

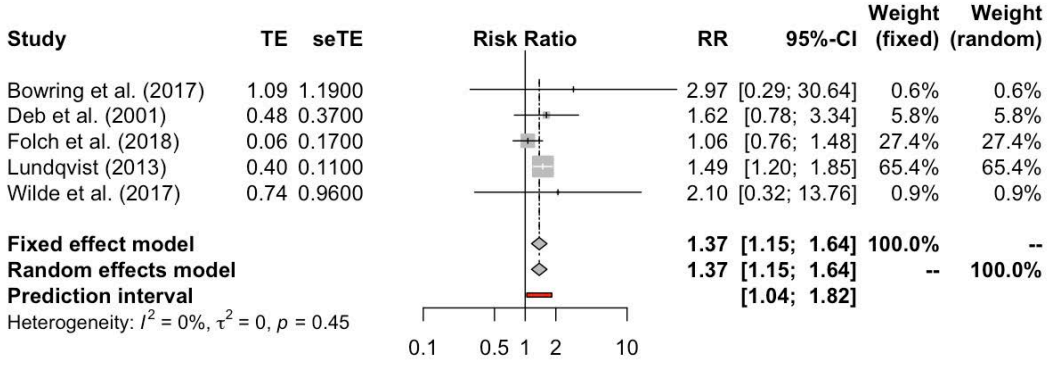
B15.1 Forest plot for all studies included for RR of mobility deficit and self-injury



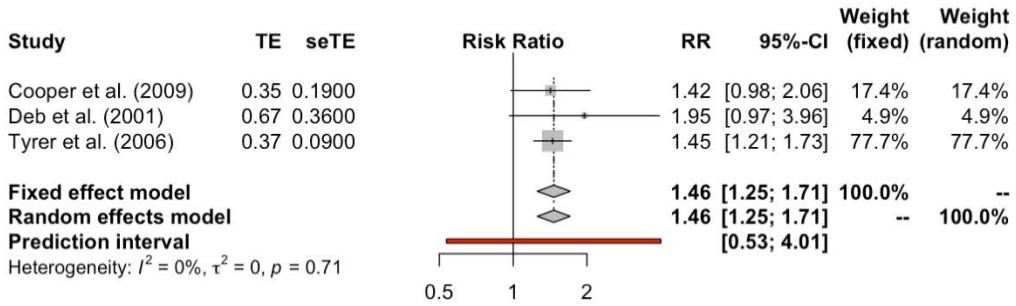
B15.2 Forest plot for all studies included for RR of mobility deficit and aggression

B16 Epilepsy

B16.1 2 Forest plot for all studies included for RR of epilepsy and self-injury

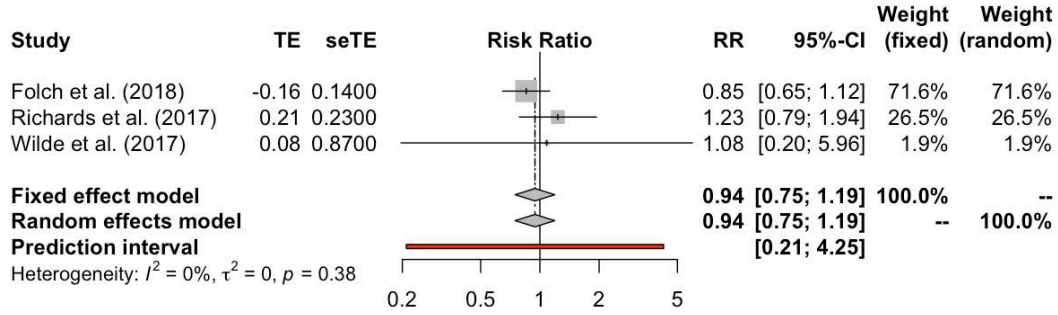


B16.2 Forest plot for all studies included for RR of epilepsy and aggression



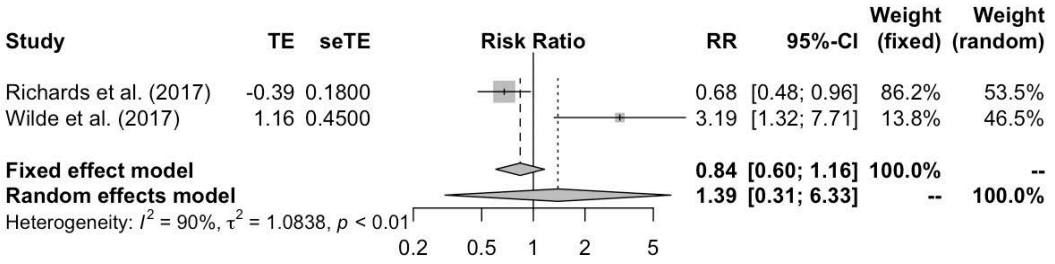
B17 Dental problems

B17.1 Forest plot for all studies included for RR of dental problems and self-injury



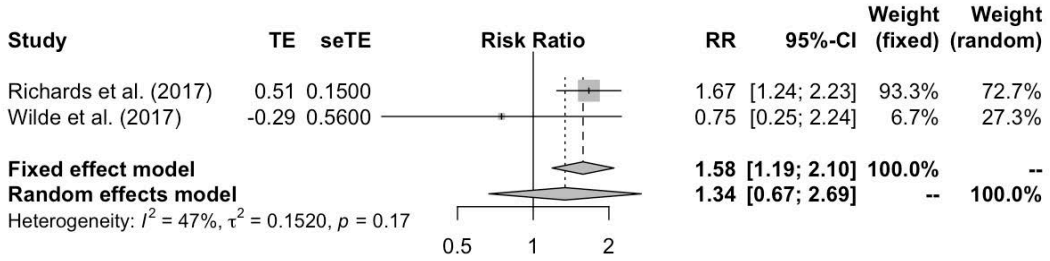
B18 Gastrointestinal problems

B18.1 Forest plot for all studies included for RR of gastrointestinal problems and self-injury



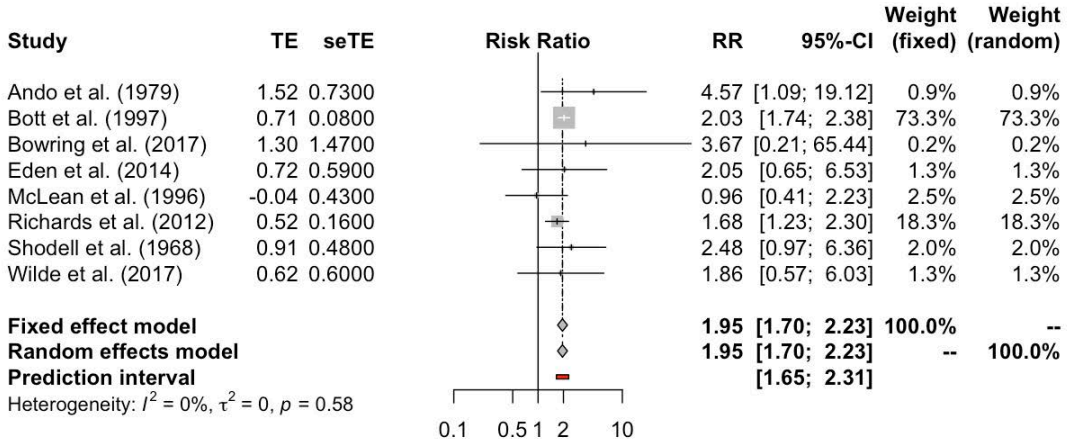
B19 Skin problems

B19.1 Forest plot for all studies included for RR of skin problems and self-injury

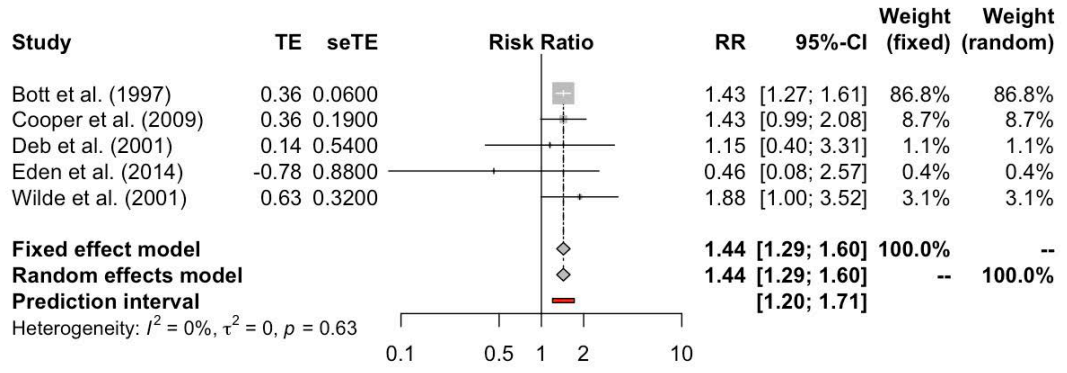


B20 Expressive communication

B20.1 Forest plot for all studies included for RR of expressive communication and self-injury



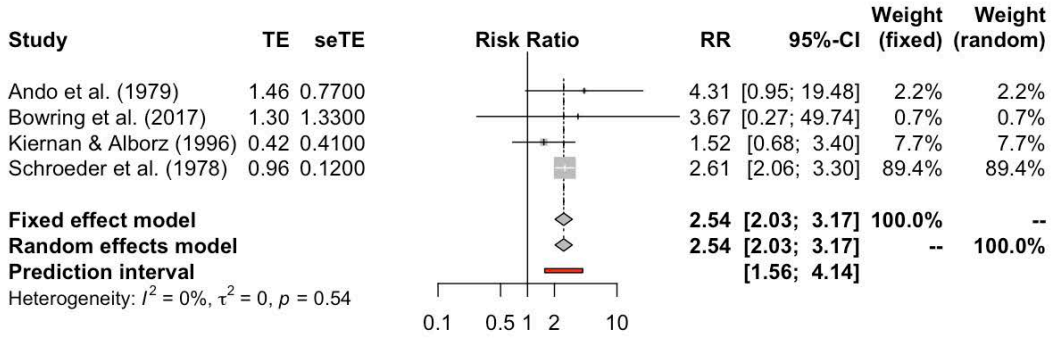
B20.2 Forest plot for all studies included for RR of expressive communication and aggression



B21 Receptive communication

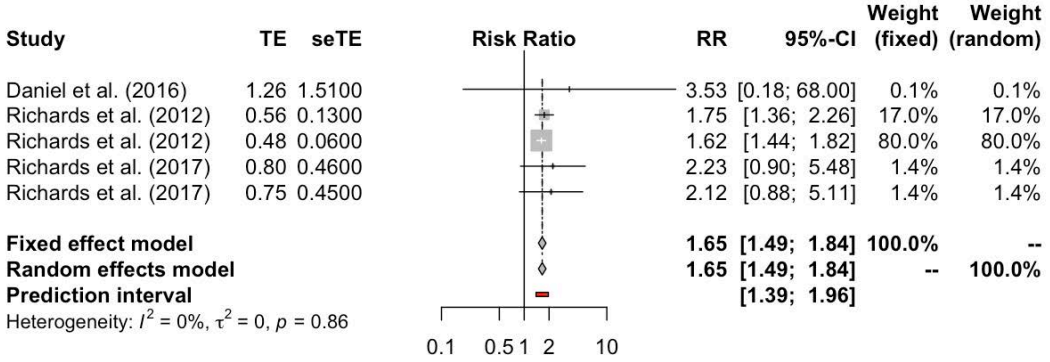
B21.1 Forest plot for all studies included for RR of receptive communication and self-injury

B22



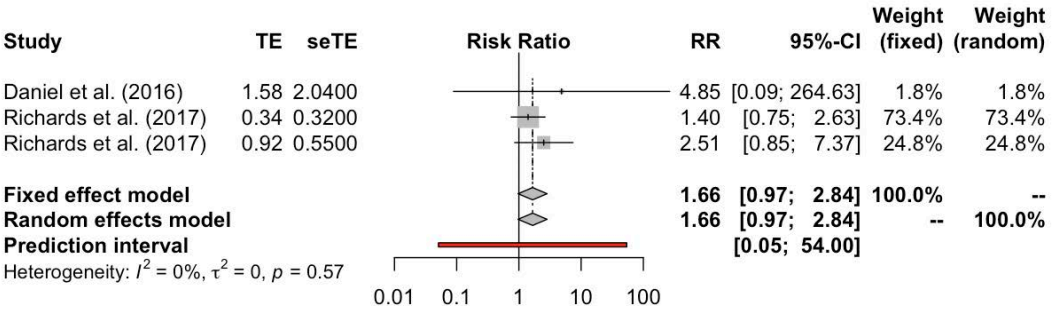
Overactivity/impulsivity

B22.1 Forest plot for all studies included for RR of overactivity/impulsivity and self-injury



B23 Repetitive behaviour

B23.1 Forest plot for all studies included for RR of repetitive behaviour and self-injury



Appendix C: HRA letter of ethical approval

Professor Chris Oliver

Professor of Neurodevelopmental Disorders

Director of the Cerebra Centre for Neurodevelopmental Disorders

School of Psychology

University of Birmingham

B15 2TT

15 August 2018

HRA and Health and Care Research Wales (HCRW) Approval Letter

Dear Professor Oliver

Study title: **The Identification of young children at highest Risk for developing Severe Challenging behaviour (i-RISC): Proof of principle and appraisal of feasibility.**

IRAS project ID:	**235418**
Protocol number:	**RG_17-182**
REC reference:	**18/NE/0249**
Sponsor	**University of Birmingham**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

How should I continue to work with participating NHS organisations in England and Wales?
You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations in England and Wales that are hosting all site activities should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of assessment*" section towards the end of this letter. You should then work with each organisation that has confirmed capacity and capability and provide clear instructions when research activities can commence.

Participating NHS organisations in England and Wales that are acting as participant identification centres **will not** be required to formally confirm capacity and capability before you may commence research activity at site. As such, you may commence the research at each organisation 35 days following sponsor provision to the site of the local information pack, so long as:

- You have contacted participating NHS organisations (see below for details)
- The NHS organisation has not provided a reason as to why they cannot participate
- The NHS organisation has not requested additional time to confirm.

You may start the research prior to the above deadline if the site positively confirms that the research may proceed.

If not already done so, you should now provide the local information pack for your study to your participating NHS organisations. A current list of R&D contacts is accessible at the NHS RD Forum website and these contacts MUST be used for this purpose. After entering your IRAS ID you will be able to access a password protected document (password: **White22**). The password is updated on a monthly basis so please obtain the relevant contact information as soon as possible; please do not hesitate to contact me should you encounter any issues.

Commencing research activities at any NHS organisation before providing them with the full local information pack and allowing them the agreed duration to opt-out, or to request additional time (unless you have received from their R&D department notification that you may commence), is a breach of the terms of HRA and HCRW Approval. Further information is provided in the "*summary of assessment*" section towards the end of this document.

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has

been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?

You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is

Name: Dr Sean Jennings

Tel: 01214158011

Email: researchgovernance@contacts.bham.ac.uk

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below. Your

IRAS project ID is **235418**. Please quote this on all correspondence.

Yours sincerely

Helen

Email: hra.approval@nhs.net

Appendix D: Parent contact letter

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098

Laura Groves: LXG502@student.bham.ac.uk, 0121 414 9775

Dear Parent/Carer,

We are writing to let you know about a new research project that will be taking place at the Cerebra Centre for Neurodevelopmental disorders at the University of Birmingham.

We have contacted you because your child attends a Special Needs School in the West Midlands**.** Your child's Head Teacher has sent out this information on our behalf. Your personal details will not be known to us unless you decide to take part in the research study and contact us directly.

The project is called 'Behaviours of children with neurodevelopmental difficulties' and aims to describe and assess different behaviours of children who are a delayed in some aspect of their development such as communication, social interaction or physical skills. The study involves the completion of a brief 10-15 minute questionnaire, focussing on a wide spectrum of behaviours that your child may or may not demonstrate.

We have enclosed an information sheet that gives you more details about what participation in this research will involve, what will happen to the information you provide and the benefits of participating. We have also enclosed a consent form, the questionnaire and two pre-paid envelopes. If you would like to participate, we would very much appreciate it if you could complete the consent form and questionnaire and return them to the research team separately in the prepaid envelopes. This is to ensure your personal identifying information, cannot be linked to your questionnaire responses, without your personal identification number.

If you have any questions / concerns about the research, please do not hesitate to contact Laura Groves.

Due to our methods of recruitment, there is a small possibility you may have already been contacted by the Cerebra Centre to take part. If this is the case, please ignore the latest invitation to participate which has been sent to you.

This research project has been approved by the North East Tyne & Wear South Research Ethics Committee and has all the necessary approvals (Health Research Authority reference number: 235418; Protocol number: RG_17 182; REC number: 18/NE/0249).

Thank you very much for your time,

Kind regards, Dr Caroline Richards, Dr Debbie Allen and Professor Chris Oliver

Appendix E: Participant Information Sheet

Behaviours of children with neurodevelopmental difficulties

Participant Information Sheet

We would like to invite you to take part in a research study being conducted at the Centre for Neurodevelopmental Disorders (CNDD) in the School of Psychology at the University of Birmingham.

Please read this information carefully before deciding whether you wish to take part in the research study. If you would like any more information about the study or you have any health and / or language difficulties which make it difficult for you to read this information please contact the research team or you can ask someone to contact the research team on your behalf (see details at the end of the information sheet).

This research project has been approved by the North East Tyne & Wear South Research Ethics Committee and has all the necessary approvals (Health Research Authority reference number: 235418; Protocol number: RG_17 182; REC number: 18/NE/0249).

What is the research study about and why is it important?

This research project, led by Professor Chris Oliver, Laura Groves and Dr Caroline Richards, is based in the Cerebra Centre for Neurodevelopmental Disorders (CNDD) in the School of Psychology at the University of Birmingham. The study aims to better understand the various behaviours of children with neurodevelopmental difficulties. We will use the information provided by parents to understand whether particular characteristics, such as age or gender, make certain behaviours more or less likely. We are also seeking to test whether the questionnaire used in this study could be useful in a clinical setting, as a screening tool.

In the future, the information obtained during this study could help us to identify ways in which we may be able prevent the onset of particular behaviours that have a negative impact on the quality of life of children with neurodevelopmental difficulties.

Why have I been invited to take part in the study?

You have been invited to take part in this study because you are the parent / carer of a child aged between 2 and 11 years old who attend one of a range of educational and health facilities / services in the West Midlands specifically for children with neurodevelopmental difficulties. You may also have received this invitation if your child is undergoing an assessment of their development or you have previously provided consent for your details to be held on the CNDD research participant database and to be contacted about future research projects. Unless your details are already held on the CNDD participant database, we do not have your contact details and we do not know your name or your child's name. The health / education facility that your child attends has sent out this information on behalf of the research team so please be assured that we do not have access to any of your personal information.

If I decide to participate in the research study, what will I be asked to do?

If you would like to participate in the research, you will be asked to sign a consent form, and provide us with a few personal details (your name, postal address, telephone number, email address, and your child's gender and age) you will then be asked to complete a short questionnaire about your child's current behaviour. This questionnaire will take between 10-15 minutes to complete.

You will be asked to complete the questionnaire and return it to the research team in a pre-paid envelope along with a completed consent form. Alternatively, you can contact a member of the research team and arrange to have the questionnaire administered over the telephone or, if you prefer, they may be able to visit you at home if you live within a reasonable travelling distance.

You will be contacted by the research team again 12 months later (Time 2) to complete the same questionnaire that you did at Time 1. This is to help the study team understand how behaviour changes over time. The research team will post the questionnaire to your home address with a pre-paid envelope for you to return the completed questionnaire and / or telephone you to administer the questionnaire on the telephone. Once again, a member of the research team may also be able to visit you at home

Who will be involved in collecting the information?

Members of the research team at the Cerebra Centre for Neurodevelopmental Disorders based at the University of Birmingham including experienced academic research staff and supervised undergraduate and postgraduate students. The research team will also include NHS Research Nurses.

What are the possible benefits of taking part in this study?

After you have completed the questionnaire, you will receive a summary report specific to your child based on your responses to the questionnaire and the opportunity to ask any questions. You will receive a similar report 12 month later, when you have completed the questionnaire for the second time.

These individualised feedback reports may be useful for you and the health and education professionals involved with your child / the child you care for to highlight particular difficulties that your child might face and identify resources that might be useful for them. We happily share a copy of this report with any health / education professionals if you request this and provide us with written consent.

What are the possible disadvantages of taking part in this study?

While the questionnaire is very brief, it includes questions about challenging behaviour which you might find upsetting. If this does happen and you feel that you do not want to continue with participation in the research, you can withdraw at any time. You will also be given the opportunity to contact the research team and discuss any concerns you have about the project at any stage during the 12-month research period who may be able to signpost you to helpful resources or professionals.

If I decide to participate, what will happen to the information I provide?

Personal identifying information, such as names, ages, addresses, telephone numbers and email addresses will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998 & 2018. Personal identifying information will be stored for the duration of the study on a password protected portable hard drive that is kept in a locked filing cabinet at the University of Birmingham.

Your completed questionnaires will be stored separately from the personal identifying information described above in a locked filing cabinet at the University of Birmingham. Your name will not appear on the completed questionnaires. Instead, each participant will be allocated a participant number and this will appear on the questionnaires instead of names. An electronic file will be created which links the participant number to the participant name which will be stored on a password protected portable hard drive kept in a locked filing cabinet at the University of Birmingham. Only the research team have access to the research project filing cabinets.

In the unlikely event that the research team have concerns about the welfare of a participant, information will be disclosed as necessary.

What would happen to the information after the project ends?

If you are not already on the existing participant database, your contact details will be destroyed at the end of the study (within 6 months of receipt of your Time 2 questionnaire or 6 months after the end of the Time data collection cut-off date) and you will not be contacted further unless your details are already stored on the CNDD database. Your anonymous completed questionnaires will be stored for up to 10 years following the end of the project.

The research team will publish the findings from the study in scientific journals and will present the results at relevant conferences and in newsletters. Any published reports which use the information you have provided us, will be completely confidential, and will never use your child's name.

If I would like to participate in the project, what should I do now?

Please remember that participation in the project is purely optional and the decision not to participate will not restrict access or affect the right to any education / health services. When you are satisfied that

you have all of the information you need to be able to decide whether or not you / the person you care for would like to take part in the study and if you decide that you would like to participate, please complete the enclosed consent form and the questionnaire and return them to us separately in the two prepaid envelope provided. This is to ensure your responses and personal details cannot be matched except by the identified number on the envelopes.

What if I change my mind about participating after I have provided consent?

Even after you have provided consent to participate in the study, you can request to be withdrawn from the study and for your research data to be destroyed without giving a reason. This will not restrict access or affect the right to any education / health services. You will have up to 6 months after completion of the Time 2 questionnaire to indicate that you would like to withdraw from the study. This is purely for practical reasons as after your personal identifying information has been destroyed, your personal details will no longer be linked to the information collected as part of this study. This means that we would no longer be able to trace the results of your assessments back to you and withdraw you from the study.

What can I do if I have any concerns about the research or there is a problem?

In the unlikely event that you have any cause for concern about any aspect of the research, in the first instance, please contact Professor Chris Oliver (Chief Investigator) on 0121 414 4909, c.oliver@bham.ac.uk, Laura Groves (Research Assistant) on 0121 414 9775, LXG502@student.bham.ac.uk, or Dr Caroline Richards (Principal Investigator) on 0121 415 8098, c.r.richards@bham.ac.uk at the University of Birmingham. If you wish to speak to someone who is independent to the research, or you have any concerns related to the research after contacting the Chief Investigator, Research Assistant or Principal Investigator, you can contact Birmingham Community Healthcare Foundation Trust team on:

Tel - 0800 917 2855

E-mail - complaints.bchc@nhs.net

Write to - Complaints Team, 3 Priestley Wharf, Holt Street, Aston, Birmingham, B7 4BN.

How do I contact the research team to find out more about the research study?

Please contact Laura Groves on:

Tel - 0121 414 9775

E- mail - LXG502@student.bham.ac.uk

Write to - Laura Groves, School of Psychology, 52 Pritchatts Road, University of Birmingham, Edgbaston, Birmingham, B15 2TT

Or Dr Caroline Richards on:

Tel - 0121 415 8098

E- mail - c.r.richards@bham.ac.uk

Write to - Caroline Richards, School of Psychology, 52 Pritchatts Road, University of Birmingham, Edgbaston, Birmingham, B15 2TT

Alternatively, you can contact Birmingham Community Healthcare Foundation Trust Customers Services Team for independent advice about the study on:

Tel - 0800 917 2855

E- mail - contact.bchc@nhs.net

Write to - Customer Services Team, The Lodge, Moseley Hall Hospital, Alcester Road, Moseley, Birmingham, B13 8JL.

The University of Birmingham is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. The University of Birmingham will keep identifiable information about you for 6 months following receipt of your Time 2 questionnaire to enable you to receive your Time 2 feedback report. If the Time 2 questionnaire isn't completed, any identifiable data will be destroyed 6 months after the data collection window.

Your rights to access, change or move your information are limited, as we need to manage information in specific ways in order for research to be reliable and accurate. If you withdraw from the study, we will keep the information we have already obtained. To safeguard your rights, we will use minimum personally-identifiable information possible.

You can find out more information about how we use your information by contacting the Data Protection Officer at the University, Mrs Carolyn Pike (OBE) legalservices@contacts.bham.ac.uk

The University of Birmingham will use your name, and contact details (and your child's age and gender) to contact you about the research study and to oversee the quality of the study. Individuals from the University of Birmingham and regulatory organisations may look at your research records check the accuracy of the research study. The only people in the University of Birmingham who will have access to the information that identifies you will be the people who need to contact you about your participation in the study or audit the data collection process.

Health and care research should serve the public interest, which means that we have to demonstrate that our research serves the interests of society as a whole. We do this by following the UK Policy Framework for Health and Social Care Research.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. Our Data Protection Officer is Mrs. Carolyn Pike OBE and you can contact them at legalservices@contacts.bham.ac.uk If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Thank you for your time.

Appendix F: Consent form

Behaviours of Children with Neurodevelopmental Difficulties

Parental Consent Form

If you are reading this information on behalf of someone you care for who is a child ages 2-11, then we would like to ask you to decide whether or not you think that it is in your child's best interests for them to participate in the study and whether you would like to provide your consent to participation on their behalf. If you would like your child/person you care for to participate in this study, please complete the consent form and return it to the research team in the prepaid envelope provided.

PART 1 **Please initial box…**

1. I confirm that I have read and understood the information sheet dated V5 18.09.18 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that participation involves completion of 1 short questionnaire now (Time 1) and that I will be contacted again in 12 months (Time 2) to complete the same questionnaire again, using the contact details I provide. ☐
3. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐
4. I understand that all information collected during the study will be confidential. Only members of the research team at the Cerebra Centre for Neurodevelopmental disorders will know who has participated in the ☐

study. All information collected during the study will be stored in locked cabinets and / or on password protected portable hard drives that only members of the research team will have access to. No names will be published in any reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 2018.

5. I understand that my contact details will only be used by the research team for the purpose of this study only.
6. I agree, on behalf of myself and the child I care for, to take part in the study 'Behaviours of Children with a Neurodevelopmental Difficulties'. ☐

Please complete the details below

Name of Parent / Carer: _____

Your relationship to the participant: _____

Signed: _____ Date: _____

Researcher name (if present)_____ Countersigned:_____

PART 2

Child's gender: Male Female

Age of Child: _____years _____months.

Address: _____

Landline Telephone Number: _____

Mobile Telephone Number: _____

Email Address: _____

Appendix G: SAD-SQ Questionnaires given to children under age 6

Child Health & Behaviour Screen

Behaviours of Children with Neurodevelopmental Difficulties

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098

Laura Groves: LXG502@student.bham.ac.uk, 0121 414 977

Today's date: ..... **Participant ID Number**:……

Child's gender: **Male** □ **Female** □ Child's Age: __(Years)___(Months)

Q1.Has any professional (e.g. doctor, geneticist, paediatrician etc.) said that the child you care for:

(Please circle your response)

Is autistic, has an autistic spectrum disorder, autistic like traits and/or features of autism? Yes / No

Has cerebral palsy or muscular dystrophy? …….. Yes / No

Has a genetic syndrome (e.g. Downs Syndrome, Fragile X, Cri du Chat etc.)?…… Yes / No

If you answered **Yes** to genetic syndrome, please state which: ……

If you answered **No** to genetic syndrome, please state the cause of the child's learning disability (if known):

……

Q2. Please name any prescribed medications the child is currently taking whilst at school:…….

……..

Q3. Please give details of any contact/visits the child has had with any health professionals

(e.g. dentists, schools nurses, psychologists etc.) in the last month regarding the child's:

Health:.....

Behaviour: ……..

Q4. Have you ever thought about seeking help regarding the child's behaviour?

Yes and have made contact □ Yes but not made contact □. No □

Q5. Please indicate the number of days that the child did not attend school in the last full term:

0-4 □ 5-9 □ 10-14 □ 15+ □

Q6 Please circle the appropriate response regarding the child's general development

Smile spontaneously	Yes/No	Turn to a voice	Yes/No
Feed self	Yes/No	Imitate speech sounds	Yes/No
Wave bye-bye	Yes/No	Say 2 words	Yes/No
Use spoon/fork	Yes/No	Point to pictures	Yes/No
Put on clothing	Yes/No	Name 1 colour	Yes/No
Grasp rattle	Yes/No	Roll over	Yes/No
Thumb and finger grasp	Yes/No	Stand holding on	Yes/No
Scribbles	Yes/No	Stand alone	Yes/No
Build a tower of two cubes	Yes/No	Run	Yes/No
Imitate drawing a vertical line	Yes/No	Jump up	Yes/No

Q7. To what extent have the following health problems affected the child in the last month?

	Severe	**Never**	**Mild**	**Moderate**
Eye problems (e.g. infections)	0	1	2	3
Ear problems (e.g. infections)	0	1	2	3
Dental problems (e.g. cavities/gum problems)	0	1	2	3
Digestive problems (e.g. reflux/stomach problems)	0	1	2	3
Skin problems (e.g. eczema/dry skin)	0	1	2	3
Respiratory problems (e.g. asthma/infections)	0	1	2	3
Epilepsy	0	1	2	3
Any other health or painful condition, (please specify)	0	1	2	3

Q8. Please circle the appropriate response to indicate whether your child is able to carry out each skill.

Is your child able to request Food?..... Yes / No

Toys?.....Yes / No

Activities?..... Yes / No

Is your child able to request help/ assistance?..... Yes / No

Is your child able to request to take a break?..... Yes / No

Is your child able to reject items/ activities?..... Yes / No

Is your child able to communicate Yes?..... Yes / No

No?..... Yes / No

Is your child able to respond appropriately to being asked to "wait"?..... Yes / No

Is your child able to respond to the following **Visual** Directions (gestures/ signs)?

Respond to their name being signalled…... Yes / No

"Come here"…….. Yes / No

"Stop"……. Yes / No

"Sit down"……. Yes / No

"Give it to me"……...…… Yes / No

"Go get…" (Familiar item)……...…… Yes / No

"Go to…" (Familiar location)……... Yes / No

"Put it back/down" …… Yes / No

"Let's go/ come with me"……...Yes / No

Is your child able to respond to the following **Verbal** Directions (spoken)?

To their name being called…….. Yes / No

"Come here"……..… Yes / No

"Stop"……..... Yes / No

"Sit down"…….. Yes / No

"Give it to me"…….. Yes / No

"Go get…" (Familiar item)…… Yes / No

"Go to…" (Familiar location)…….. Yes / No

"Put it back/ down"……... Yes / No

"Let's go/ come with me"……. Yes / No

Is your child able to react appropriately to changing activities?..... Yes / No

Is your child able to follow a visual schedule?..... . Yes / No

Q9. Please circle your response to each question.

Does the child you care for:	**How frequently does this problem occur from never (0) to very often (4)?**					**How difficult is it to manage this problem from not difficult (0) to extremely difficult (4)?**					**How concerned are you about this problem from not at all concerned (0) to extremely concerned (4)?**				
Show repetitive movements?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show obsessions and rituals?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show destruction of property (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show self injury (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show emotional outbursts (e.g. excessive emotions including anger or distress etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult to wait?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Act as if driven by a motor?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Want things immediately?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult holding still?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4

Appendix H: SAD-SQ Questionnaires given to children aged 6 and over

Child Health & Behaviour Screen

Behaviours of Children with Neurodevelopmental Difficulties

Dr Caroline Richards: c.r.richards@bham.ac.uk, 0121 415 8098

Laura Groves: LXG502@student.bham.ac.uk, 0121 414 977

Today's date: ..... **Participant ID Number**:……

Child's gender: **Male** □ **Female** □. Child's Age: __(Years)___(Months)

Q1.Has any professional (e.g. doctor, geneticist, paediatrician etc.) said that the child you care for:

(Please circle your response)

Is autistic, has an autistic spectrum disorder, autistic like traits and/or features of autism? Yes / No

Has cerebral palsy or muscular dystrophy? …….. Yes / No

Has a genetic syndrome (e.g. Downs Syndrome, Fragile X, Cri du Chat etc.)?…… Yes / N

If you answered **Yes** to genetic syndrome, please state which: ……

If you answered **No** to genetic syndrome, please state the cause of the child's learning disability (if known):

……

Q2. Please name any prescribed medications the child is currently taking whilst at school:…….

……..

Q3. Please give details of any contact/visits the child has had with any health professionals

(e.g. dentists, schools nurses, psychologists etc.) in the last month regarding the child's:

Health:.....

Behaviour:……..

Q4. Have you ever thought about seeking help with the child's behaviour?

(Please circle your response)

Yes and have made contact □ Yes but not made contact □ No □

Q5. Please indicate the number of days that the child you care for did not attend school in the last full term. (Please circle your response)

0-4 □ 5-9 □ 10-14 □ 15+ □

Q6. Please circle the appropriate response regarding the child's general development

Walk without help	1…Not at all	2…Not upstairs	3…Upstairs and elsewhere
Feed self	1…Not at all	2…With help	3…Without help
Wash self	1…Not at all	2…With help	3…Without help
Dress self	1…Not at all	2…With help	3…Without help
Wetting (days)	1…Frequently	2…Occasionally	3…Never
Soiling (days)	1…Frequently	2…Occasionally	3…Never
Reads	1…Nothing	2…A little	3…Newspapers and/or books
Writes	1…Nothing	2…A little	3…Own correspondence
Counts	1…Nothing	2…A little	3…Understands money values
Speech	1…Never a word	2…Odd words only	3…Sentences and normal
Vision	1…Blind or almost	2…Poor	3…Normal
Hearing	1…Deaf or almost	2…Poor	3…Normal

Q7. To what extent have the following health problems affected the child in the last month?

(Please circle your response)

	Never	**Mild**	**Moderate**	**Severe**
Eye problems (e.g. infections)	0	1	2	3
Ear problems (e.g. infections)	0	1	2	3
Dental problems (e.g. cavities/gum problems)	0	1	2	3
Digestive problems (e.g. reflux/stomach problems)	0	1	2	3
Skin problems (e.g. eczema/dry skin)	0	1	2	3
Respiratory problems (e.g. asthma/infections)	0	1	2	3
Epilepsy	0	1	2	3
Any other health or painful condition	0	1	2	3

Q8. Please circle the appropriate response to indicate whether your child is able to carry out

each skill.

Is your child able to request Food?..... Yes / No

Toys?..... Yes / No

Activities?..... Yes / No

Is your child able to request help/ assistance?..... Yes / No

Is your child able to request to take a break?..... Yes / No

Is your child able to reject items/ activities?..... Yes / No

Is your child able to communicate Yes?..... Yes / No

No?... ..... Yes / No

Is your child able to respond appropriately to being asked to "wait"?..... Yes / No

Is your child able to respond to the following **Visual** Directions (gestures/ signs)?

Respond to their name being signalled….. Yes / No

"Come here"…….. Yes / No

"Stop"……. Yes / No

"Sit down"……. Yes / No

"Give it to me"……..…… Yes / No

"Go get…" (Familiar item)……..…… Yes / No

"Go to…" (Familiar location)……... Yes / No

"Put it back/down" …… Yes / No

"Let's go/ come with me"……... Yes / No

Is your child able to respond to the following **Verbal** Directions (spoken)?

To their name being called…….. Yes / No

"Come here"……..… Yes / No

"Stop"……..... Yes / No

"Sit down"…….. Yes / No

"Give it to me"…….. Yes / No

"Go get…" (Familiar item)…… Yes / No

"Go to…" (Familiar location)…….. Yes / No

"Put it back/ down"……... Yes / No

"Let's go/ come with me"……. Yes / No

Is your child able to react appropriately to changing activities?..... Yes / No

Is your child able to follow a visual schedule?..... . Yes / No

Q9. Please circle your response to each question.

Does the child you care for:	**How frequently does this problem occur from never (0) to very often (4)?**					**How difficult is it to manage this problem from not difficult (0) to extremely difficult (4)?**					**How concerned are you about this problem from not at all concerned (0) to extremely concerned (4)?**				
Show repetitive movements?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show obsessions and rituals?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show destruction of property (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show self injury (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Show emotional outbursts (e.g. excessive emotions including anger or distress etc.?)	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult to wait?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Act as if driven by a motor?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Want things immediately?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4
Find it difficult holding still?	0	1	2	3	4	0	1	2	3	4	0	1	2	3	4